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Original Paper

Accuracy and Completeness of the Registration of Childhood Leukaemia in The Netherlands, 1989–1992

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In The Netherlands, childhood leukaemia is recorded by the Dutch Childhood Leukaemia Study Group (DCLSG, set up in 1972) and by nine regional cancer registries which together form the Netherlands Cancer Registry (NCR, set up in 1989). The data files from the incidence years 1989–1992 of the two registries were linked in order to evaluate accuracy and completeness and to calculate and equalise the incidence rates for childhood leukaemia in The Netherlands. Unlinked records or records with disagreements (birth date, sex, type of leukaemia and incidence date) were checked by the DCLSG and by the regional cancer registries. The DCLSG recorded 431 cases of childhood leukaemia, while the NCR recorded 434 cases. After record linkage and review of the cases, it was concluded the 445 records should have been recorded as childhood leukaemia. The NCR had recorded 425 of the 445 correct cases (95.5%), but had missed 20 cases (4.5%). The DCLSG had recorded 431 of the 445 correct cases (96.9%) and had missed 14 cases (3.1%). In addition, the NCR had recorded 9 cases incorrectly as childhood leukaemia. Part of the disagreement was caused by differences in coding rules (definition of non-Hodgkin's lymphoma (NHL) and the myelodysplastic syndrome versus leukaemia). It could be concluded that the quality and completeness of the two registries was very high. Regular comparison of the recorded data will help to reveal the inherently problematic disagreement between definitions and coding. © **1997 Elsevier Science Ltd.**

Key words: childhood leukaemia, completeness, cancer registry, The Netherlands

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INTRODUCTION

IN ORDER for a cancer registry to be reliable and useful, it is essential that the data are complete and valid. Investigation of trends in time and of possible clusters in location and time may be biased by low validity and/or low completeness of the cancer registry data. This is especially true for relatively rare malignancies, such as childhood leukaemia. The latter malignancy has frequency been the subject of investigations of trends and possible clusters [1–3].

In The Netherlands, childhood leukaemia is recorded by the Dutch Childhood Leukaemia Study Group (DCLSG, set up in 1972) and by the Netherlands Cancer Registry (NCR, set up in 1989) which consists of nine regional cancer registries. Owing to the fact that the two registries had been requested to submit incidence data to the International Agency for Research on Cancer for inclusion in the second volume of the International Incidence of Childhood Cancer, data files from the incidence years 1989–1992 were linked:

—to assess the accuracy and completeness of both registries; and

—to calculate and equalise the incidence rates for childhood leukaemia in The Netherlands. An analysis of these data is presented here.

MATERIALS AND METHODS

Recording procedures of the two cancer registries

The data of the NCR are collected by the nine regional cancer registries (RCRs) [4]. In each region, the RCR receives lists of newly diagnosed cases on a regular basis from pathology and haematology departments, the medical

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records departments and some radiotherapy institutes. Following this notification, trained registration personnel from the cancer registries abstract the relevant information for the cancer registry from the medical records at the hospital concerned. After checks for inconsistencies and duplicate records, the data are entered into the national database. The NCR has been recording data on newly diagnosed malignancies since 1989.

The DCLSG is the central reference centre for the diagnosis and treatment of childhood leukaemia in The Netherlands. Since 1972, the DCLSG laboratories have been assessing the cytomorphology of each (suspected) case of childhood leukaemia and since 1978 also the immunophenotype. In addition, all clinically relevant data at diagnosis, during treatment and during long-term follow-up, are recorded by a network of data managers according to DCLSG protocols. A previous valuation of the completeness of the DCLSG registry has shown that 95.4–99.9% of all the eligible cases of childhood leukaemia in The Netherlands were recorded by the DCLSG in the period 1973–1980 [5].

Owing to privacy regulations, NCR and the DCLSG registry do not receive any notification by means of death certificates.

Record linkage and elucidation of disagreements

Before submitting data for publication in the second volume of the International Incidence of Childhood Cancer, computer files of new cases of childhood leukaemia diagnosed from 1989 to 1992 inclusive were linked using date of birth and sex as the identifiers. The correctness of the matching records was checked using the items data of incidence, type of leukaemia and municipality. Any unlinked records or records with disagreements were checked by the DCLSG and the regional cancer registries with respect to the accuracy of the identifying information, date of incidence (>1 month) and type of leukaemia. The responses of both registries were used to determine the information that was most probably coded correctly.

RESULTS

In the period of investigation (1989–1992 inclusive), the NCR had recorded 434 cases of childhood leukaemia, while the DCLSG had recorded 431 cases. Linkage was achieved for 397 records. There were eight matches with disagreements in sex or date of birth. Six DCLSG records could be linked to NCR cases recorded as non-Hodgkin's lymphoma (NHL). Therefore, a total of 411 records could be linked (Figure 1). Also, three duplicated records were detected in the NCR. Major disagreements were found in 42 records

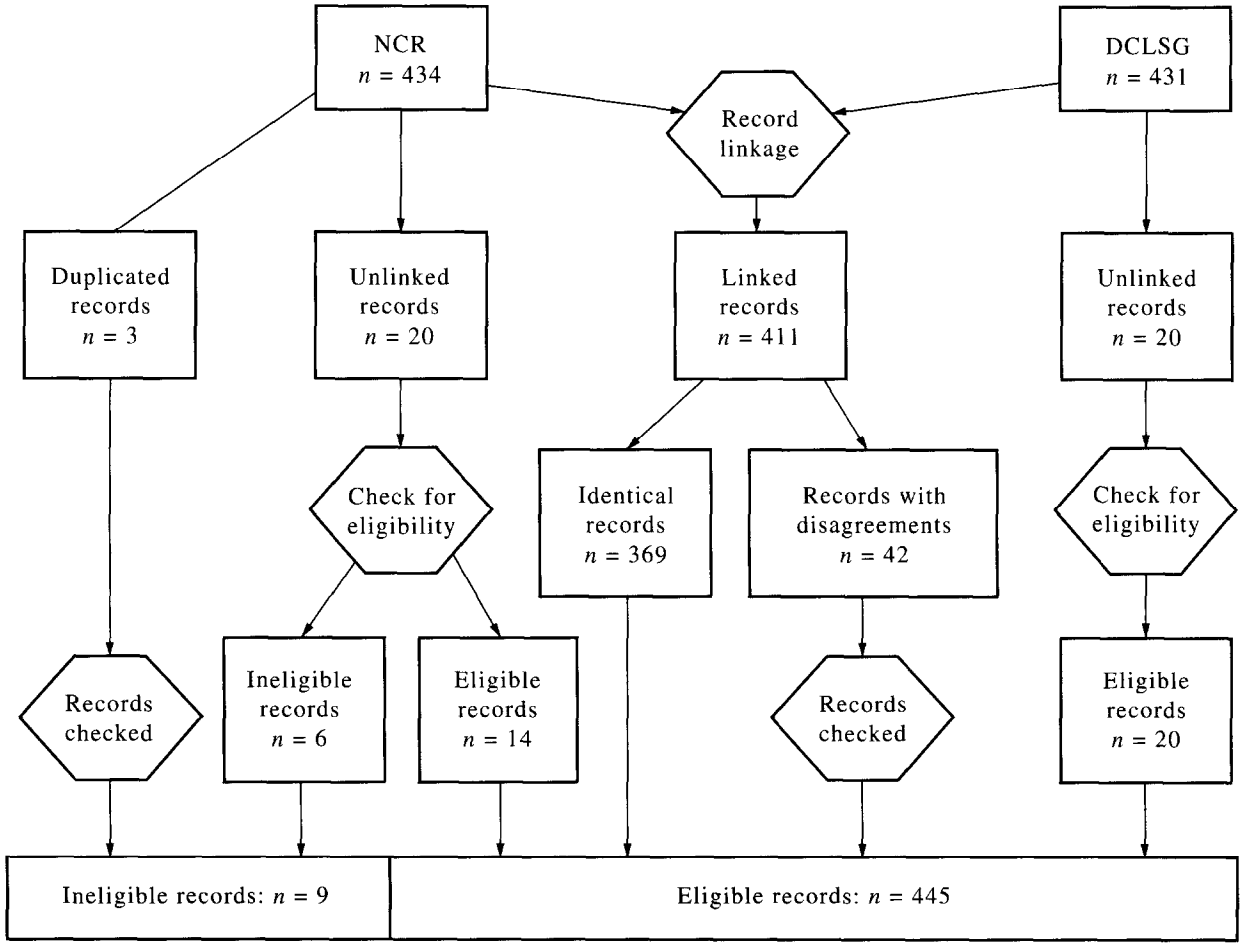

Figure 1. Diagram of record linkage, checks for accuracy and eligibility of the files of The Netherlands Cancer Registry (NCR) and the Dutch Childhood Leukaemia Study Group (DCLSG).

Table 1. Absolute number and proportion of disagreements between the NCR and DCLSG records

Item	Disagreements NCR (n = 434)		DCLSG (n = 431)	
	n	%	n	%
Date of birth	2	0.5%	3	0.7%
Sex	3	0.7%	2	0.5%
Date of incidence*	11	2.5%	6	1.4%
Subtype of leukaemia†	8	1.8%	3	0.7%

*Disagreement > 1 month. †Including malignant versus non-malignant, excluding the coding differences with respect to leukaemia/NHL (n = 4) and leukaemia/MDS (n = 5).

with respect to date of birth, sex, date of incidence or the subtype of leukaemia. No linkage was possible for 20 records from the DCLSG and for 20 records from the NCR.

After reviewing the cases, it was concluded that 445 records should have been recorded as childhood leukaemia. The NCR had recorded 425 of the 445 eligible cases (95.5%), but had missed 20 cases (4.5%). The DCLSG had recorded 431 of the 445 eligible cases (96.9%), but had missed 14 cases (3.1%). In addition, the NCR had recorded 9 cases that were not eligible.

In the 20 cases unknown to the NCR, notification had been received in the meantime by a regional cancer registry in 8 cases, but the data had not yet been submitted to the NCR. In one patient, only the first malignancy (NHL) had been recorded by the NCR, whereas the second primary (leukaemia) had not. The reasons why the other cases had not been recorded by the NCR were unknown.

Nine cases from the NCR were ineligible for different reasons. Three cases were duplicates (one caused by an error in the date of birth, the other two by disagreements in the family name), four cases were prevalent cases (diagnosed before 1989; this was detected after reviewing the clinical files), one case was not a Dutch resident, and one case did not actually have a malignancy. In total, the DCLSG had not recorded 14 eligible cases. The DCLSG had recorded 5 cases under the Myelodysplastic Syndrome (MDS). These cases had undergone progression to leukaemia, but had not been included in the incidence rates. One other case had been recorded as NHL. A total of 8 cases had not been reported to the DCLSG: one patient had been treated by a haematologist (not a paediatrician), 2 patients had died shortly after diagnosis and 5 cases had been overlooked for unknown reasons.

The NCR had recorded 4 cases as NHL, using the diagnostic definitions for adult leukaemia and NHL. In the framework of this study, it was considered that these cases should be recorded as leukaemia. However, as the cases were coded correctly according to the NCR rules, they were not regarded as errors. The disagreements with respect to date of birth, sex, date of incidence (> 1 month) and type of leukaemia are summarised in Table 1. In general, the proportion of errors was small.

Based on this study, combined incidence rates of childhood leukaemia were calculated for The Netherlands over the period 1989–1992 (see Table 2). There were 243 male cases of childhood leukaemia and 202 female cases. The annual age-adjusted incidence rate (according to the World Standard Population [6]) was estimated to be 4.5 per 100 000 males and 3.9 per 100 000 females. Using the original NCR data, the age-adjusted rates were estimated to be 4.3 per 100 000 males and 3.8 per 100 000 females; using the original DCLSG data, these rates were 4.3 per 100 000 males and 3.9 per 100 000 females.

DISCUSSION

Completeness of case ascertainment with respect to the registration of childhood leukaemia in The Netherlands was very high: the NCR was 95.5% complete, while the DCLSG was 96.9% complete. Part of the incompleteness of the NCR could be explained by a delay in submitting data to the NCR. It cannot be excluded that cases were missed by both registries. However, because of good access to specialised care in The Netherlands and active registration procedures, we assume that this number was very low. This

Table 2. Absolute numbers and incidence rates (n/100 000) of childhood leukaemia in The Netherlands, 1989–1992, according to the combined data from the Netherlands Cancer Registry and the Dutch Childhood Leukaemia Study Group

Sex and morphological type	Total	Age groups (years) 0	1–4	5–9	10–14	Crude rate	WSR*	Cumulative risk 0–14 years (%)
Males								
Acute lymphoid leukaemia	191	5	91	65	30	3.4	3.6	0.051
Acute non-lymphocytic leukaemia	32	5	12	9	6	0.6	0.6	0.008
Acute unspecified leukaemia	13	–	7	2	4	0.2	0.2	0.003
Chronic myeloid leukaemia	6	2	1	1	2	0.1	0.1	0.002
Other lymphoid leukaemia	1			1		0.0	0.0	0.000
Total	243	12	111	78	42	4.3	4.5	0.065
Females								
Acute lymphoid leukaemia	154	4	86	37	27	2.9	3.0	0.043
Acute non-lymphocytic leukaemia	29	6	3	5	15	0.5	0.5	0.008
Acute unspecified leukaemia	17	3	11	2	1	0.3	0.3	0.005
Chronic myeloid leukaemia	2		1	1		0.0	0.0	0.001
Other lymphoid leukaemia								
Total	202	13	101	45	43	3.8	3.9	0.056

*Incidence rates standardised for differences in age distribution using the World Standard Population.

is supported by the calculated estimate of missed cases using capture–recapture methods [7]. Assuming that the data collection procedures followed by the two registries were independent, less than one case would have been missed by both registries.

The estimates of completeness of the NCR are in agreement with those of other studies [7, 8]. The completeness of the IKL cancer registry (Maastricht) for all malignancies was estimated to be 96.2% in the period 1988–1990 [8]. Using the capture–recapture method, the completeness of case ascertainment for haematological malignancies in three Dutch regional cancer registries was estimated to be 95.2% [7].

The estimate of completeness of the DCLSG is very high and proves that this study group is widely supported by Dutch paediatricians. During the period 1973–1979, the completeness of the DCLSG registry was estimated to be 95.4–99.9% [5]; the current estimate is within the same range.

Using the capture–recapture method, the completeness of case ascertainment of the Ontario cancer registry for haematological malignancies was estimated to be 95% [9]. The completeness of Hodgkin's disease in the cancer registry of England and Wales was estimated to be 90% (and 93% in the regional registries of England and Wales) [10]. For all malignancies, completeness was estimated to be 94% in north-west England [11], 97.8% in Denmark [12] and 96% in Saarland, Germany [13]. The results of the NCR and the DCLSG are within the same range as those of the aforementioned registries.

The number of errors in both registries was small and they were based, to a large extent, on differences in coding rules. Furthermore, the DCLSG registry is patient-based, while the NCR is tumour-based. However, in future, any children primarily recorded as non-leukaemic patients who progress to leukaemia will be recorded as such by the DCLSG and their data will be included in the calculation of incidence rates.

The DCLSG has adopted international rules to differentiate between leukaemia and NHL in children. The DCLSG uses the proportion of blasts on the bone marrow slide to distinguish between leukaemia and NHL: $\geq 25\%$ blasts is considered to be leukaemia, $<25\%$ NHL [14]. Since 1994, a panel of pathologists have been serving as a central review committee for all NHL cases recorded by the DCLSG. It is expected that this will reduce the number of disagreements about pathology and haematology specimens. In previous years (1973–1985), a double diagnosis thus appeared to be given to 5% of all cases with acute lymphocytic leukaemia and to 19% of cases with NHL [15].

In view of the high quality of both registries, it may seem superfluous to keep more than one. However, the number of children with a diagnosis of leukaemia forms only a small proportion of the number of malignancies recorded by the NCR. If the NCR were to stop recording childhood leukaemia, this would interfere with (childhood) cancer registration in general. The DCLSG laboratory plays an important role in confirming the diagnosis and determining the type of leukaemia. In addition, the DCLSG also designs, implements and evaluates the results of treatment according to DCLSG protocols, so it also records recurrences and side-effects of treatment. In addition, the DCLSG covers a longer calendar period and is used as a basis for epidemiological studies [1].

The longstanding expertise of the DCLSG laboratory also provides a solid base for the NCR. Regular comparisons between registries, along the same lines, can yield valuable information with respect to the quality of the data. The fact that both registries are reliable and complete forms a good starting point for clinical and epidemiological studies.

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