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Under-notification of giardiasis in Auckland, New Zealand: a capture–recapture estimation

M. E. HOQUE[1]*, V. T. HOPE[1], R. SCRAGG[1] AND J. GRAHAM[2]

[1] *Section of Epidemiology & Biostatistics, School of Population Health, The University of Auckland, New Zealand*
[2] *Auckland Regional Public Health Services, New Zealand*

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SUMMARY

Estimation of the degree of undercount is important for disease surveillance. Capture–recapture techniques are now being used to evaluate the completeness of disease ascertainment. This study estimated the level of under-notification of giardiasis in the Auckland adult population using a capture–recapture method. Two independent datasets of giardiasis cases ⩾15 years were generated from the 1998–1999 Auckland Giardiasis Study (AGS) case database and cases notified to Auckland Regional Public Health Services (ARPHS) for the same period of time. Cases were matched and under-notification was estimated using a two-sample capture–recapture method. During the 12-month period, 199 cases participated in the AGS and 413 cases were notified to ARPHS. The capture–recapture calculation indicated that only 49% of cases were notified. Under-notification by a factor of 2 obscures the true burden of giardiasis. Socio-economic conditions and water quality may influence disease notification inversely. Capture–recapture techniques are useful in evaluating the completeness of surveillance.

INTRODUCTION

Population-based morbidity data are essential in monitoring disease trends and in generating hypotheses on the causation of disease. They play an important role in disease surveillance and service planning. A high coverage of incident cases is required for proper interpretation of data. However, a complete measurement of disease incidence is very difficult to achieve. A critical and important component of disease monitoring is the knowledge of the degree of undercount which has so far enjoyed little attention [1, 2]. This under-ascertainment may lead to underestimation of absolute rates and may bias time, subgroup and geographical comparisons when it varies differently by these factors. Improvement in the accuracy of rates can be achieved by correcting them for the level of ascertainment. Therefore, ascertainment level has been considered a primary determinant in the calculation of a rate [2]. Under-ascertainment can be controlled by prevention of undercount, and/or by re-estimation and correction [3]. However, a widely used method in epidemiology is to estimate the level of undercounting using capture–recapture methods [4–6]. Capture–recapture is an indirect estimate based on the degree of overlap between two or more separate samples of the population under study [5]. Such approaches are attempted to avoid an exhaustive and expensive complete survey or census and to evaluate the likely completeness of any survey or census [7].

The capture–recapture method was used originally in ecology by Peterson (1894) and Dahl (1917) to estimate the size of fish populations [5]. Later, Lincoln

* Author for correspondence: Dr M. E. Hoque, Section of Epidemiology & Biostatistics, School of Population Health, The University of Auckland, Private Bag 92019, Auckland, New Zealand. (Email: e.hoque@auckland.ac.nz)

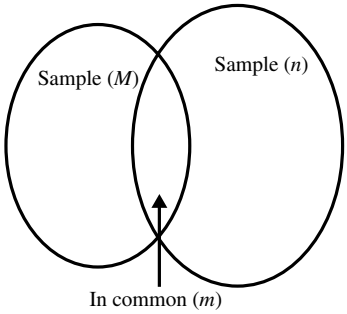

Fig. 1. Capture–mark–recapture methods.

(1930) used it to estimate the size of a duck population [8]. Application of this method to human health was first reported in India [9] and in the United States [10] in 1949 to estimate birth and death rates using census data. Medline search for relevant articles suggests that the use of capture–recapture techniques in the estimation of, or adjustment for, the extent of incomplete ascertainment has increased in recent years. These techniques have been applied in addressing the incompleteness of data collected on such diverse health conditions as birth defects, cancer mortality, diabetes, drug addiction, foetal alcohol syndrome and traffic accidents [11–19]. However, the use of capture–recapture methods in the estimation of under-ascertainment of infectious disease incidence is very limited, although, they would be potentially useful in investigating infections with mild to moderate symptoms or asymptomatic cases as they are more likely to be under-reported than severe disease. Recently, some successful attempts have been made to estimate and monitor the degree of under-reporting in sexually transmitted diseases (STD) and HIV/AIDS by using capture–recapture methodology [20–22]. To our knowledge, no attempt has so far been made to use capture–recapture methods to assess the actual number of diagnosed cases of giardiasis in the community. We have used two-sample capture–recapture methodology to estimate the extent of under-notification of giardiasis in the Auckland population aged 15 years or over ($\geqslant$15 years).

METHODS

The simplest capture–recapture model is the so-called two-sample model [23], used to estimate the total number of cases in the target population. The first sample provides the individuals for marking who are 'returned' to the population, while the second sample provides the recaptures (Fig. 1) Using the numbers of individuals found in both samples (the recaptures) and the numbers found in each sample, it is possible to estimate the number not found in either sample, thus providing an estimate of the total cases. Some key assumptions are required for this estimate to be valid: there is no change to the population during the investigation (the population is closed); there is no loss of identified cases (individuals can be matched from capture to recapture); each individual has the same chance of being included in either sample; and the two samples are independent [5].

Demographic information for all giardiasis cases aged $\geqslant$15 years reported from the Auckland Region for the period of July 1998 to June 1999 were accessed from the disease notification database of the Auckland Regional Public Health Services (ARPHS), previously known as Auckland Healthcare. Similarly, cases recruited during the Auckland Giardiasis Study (AGS) of adults aged $\geqslant$15 years for the period July 1998 to June 1999 [24], were accessed from the study data bank. Giardiasis cases mostly present with mild to moderate symptoms, many may remain asymptomatic and under-representation or under-diagnosis was reported in some ethnic groups [25]. This may culminate into positive dependence [26], however, no outbreak was reported during the study period to reason for any over-representation. Data were collected exclusively from General Practitioners (GPs) and confirmed by stool tested at one of the two diagnostic laboratories, where the chances of duplication and, therefore, negative dependence were minimal [27].

Data collected from the AGS [24] and the ARPHS notifications database were auto-geocoded using the residential addresses of patients and ArcView GIS software. This gave x,y-coordinates allowing a point of address to be marked on a map of the Auckland Region. Addresses that did not automatically geocode were done manually. These were then overlaid on a pre-existing map by drinking-water zones in Auckland regions. Drinking-water zones were used as a proxy to the Auckland Metropolitan area. Using an estimated population for each zone and the number of cases, a rate of illness was calculated. Age-specific rates for study cases and notified patients were calculated and compared. These sets of data were further analysed to detect the proportion of under-reported giardiasis cases in the Auckland Region using the two-sample capture–recapture method (Chapman estimator) [28]:

$$N = \frac{(M+1)(n+1)}{(m+1)} - 1,$$

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Table 1. *Age distribution of notified and study cases by gender*

Age group (years)	Notified cases			Study cases		
	Male No. (%)	Female No. (%)	Total No. (%)	Male No. (%)	Female No. (%)	Total No. (%)
15–24	10 (5·1)	20 (9·2)	30 (7·3)	7 (8·5)	5 (4·3)	12 (6·0)
25–34	58 (29·6)	62 (28·6)	120 (29·1)	17 (20·7)	37 (31·6)	54 (27·1)
35–44	73 (37·2)	68 (31·3)	141 (34·1)	31 (37·8)	43 (36·7)	74 (37·2)
45–54	36 (18·4)	26 (12·0)	62 (15·0)	18 (22·0)	17 (14·5)	35 (17·6)
55–64	12 (6·1)	24 (11·1)	36 (8·7)	6 (7·3)	13 (11·1)	19 (9·6)
65–74	4 (2·1)	13 (6·0)	17 (4·1)	3 (3·7)	1 (0·9)	4 (2·0)
75+	3 (1·5)	4 (1·8)	7 (1·7)	0 (0·0)	1 (0·9)	1 (0·5)
Total	196 (47·5)	217 (52·5)	413 (100·0)	82 (41·2)	117 (58·8)	199 (100·0)

Table 2. *Infection rates of notified and study cases and participation rates of study cases by area of residence*

Area of residence	Population† No. (⩾15 yr)	Notified cases‡			Study cases			Participation (%) $n/M \times 100$
		M	%	Cases/10^5	n	%	Cases/10^5	
Central Auckland	270 195	158	38·5	58·5	71	35·7	26·3	44·9
North Auckland	160 881	109	26·6	67·8	62	31·2	38·5	56·9
West Auckland	120 168	42	10·3	35·0	19	9·5	15·8	45·2
South Auckland	214 542	101	24·6	47·1	47	23·6	21·9	46·5
Total	765 786	410	100·0	53·5	199	100·0	26·0	48·5

† NZ census 1996, aged ⩾15 years.
‡ Auckland Regional Public Health Services notification (3 un-geocoded cases excluded).

where N is the estimate of the total number of cases, M is the number of cases identified in the ARPHS notifications, n is the number of cases identified by the AGS [24], and m is the number of cases identified by both the samples.

RESULTS

During a 12-month period (July 1998 to June 1999), 413 cases of giardiasis aged ⩾15 years notified to ARPHS were accessed to create an independent dataset. Collection of the second set of data involved voluntary referral of giardiasis patients by GPs to the AGS team. Over 200 laboratory-diagnosed giardiasis cases, where subjects met the entry criterion of age (⩾15 years), were referred to the study by GPs and all were interviewed. One case, for whom the questionnaire was incomplete, was excluded. Study data on the 199 cases interviewed were analysed to estimate under-notification.

Study participants were >1·4 times more likely to be female than male ($\chi^2 = 5{\cdot}92$, $P < 0{\cdot}05$) whereas the frequency of notified cases in females was 1·1 times higher than for males ($\chi^2 = 1{\cdot}07$, $P = 0{\cdot}30$). Approximately 52% notified cases and 59% study cases were females compared to 48% and 41% respectively for males. Data on age were collapsed into 10-year bands and compared by gender. The largest group was those aged 35–44 years, although the combined age groups of 25–34 and 35–44 years comprised 63% (Table 1). Age-group distribution between the cases was not significantly different ($\chi^2 = 4{\cdot}58$, $P = 0{\cdot}47$).

All the cases were grouped by place of residence into four census zones in Greater Auckland; namely, Central, North, West and South Auckland. The area of residence for three notified cases was not available and they were excluded from further analysis. The highest proportion of study (35·7%) and notified (38·5%) samples came from Central Auckland while West Auckland was the least represented (Table 2). When notified cases were compared with study cases, the study cases were slightly higher in proportion in North Auckland. However, South and West

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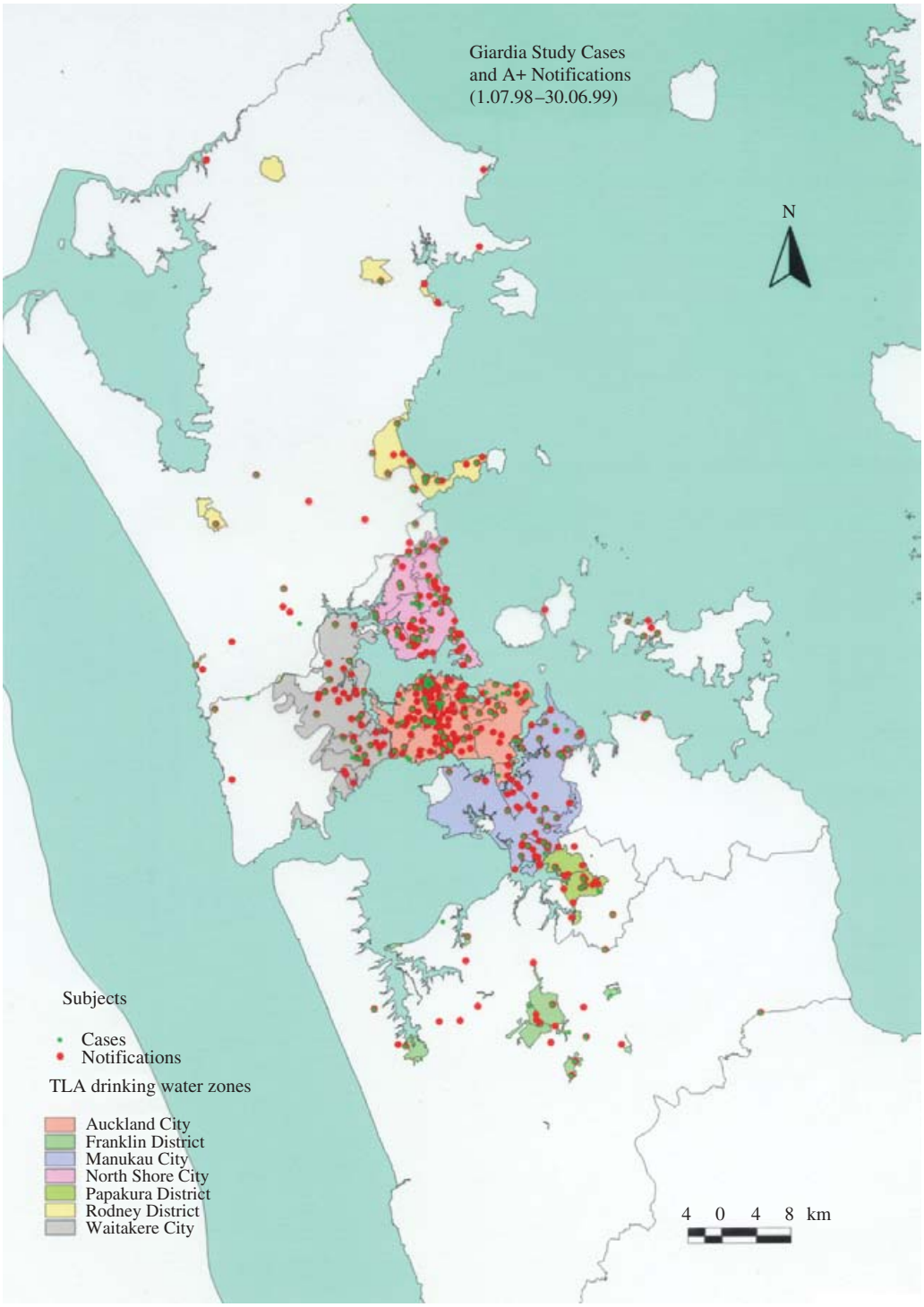

Fig. 2. Mapping of *Giardia* cases in Auckland (Auckland Giardiasis Study and Auckland Regional Public Health Services notifications) by drinking-water zones.

Auckland had almost identical proportions in both categories. The study cases and notified cases were distributed evenly by area of residence ($\chi^2 = 1{\cdot}41$, $P = 0{\cdot}70$). When incidence rates were compared, the highest rates in both categories of cases were from North Auckland and the lowest from West Auckland. Study participation rates for all regions of Auckland combined were 48·5% of notified cases.

The areas of residence of 199 study cases along with those of the 410 cases notified to ARPHS were

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Table 3. *Distribution of cases and estimation of expected notifications by area of residence*

Area of residence in Auckland	Population* P No.	No. of cases observed: Notified† M No.	Study‡ n No.	Common m No. (%)	Total cases identified $E=(n-m)+M$§ No.	% of under-notification $(1-M/E)\times 100$ %	Cases/100 000: $M/P\times 10^5$ Current	Cases/100 000: $E/P\times 10^5$ Expected
Central Auckland	270 195	158	71	31 (43·7)	198	20·2	58·5	73·3
North Auckland	160 881	109	62	37 (59·7)	134	18·7	67·8	83·3
West Auckland	120 168	42	19	8 (42·1)	53	20·8	35·0	44·1
South Auckland	214 542	101	47	22 (46·8)	126	19·8	47·1	58·7
Total	765 786	410	199	98 (49·3)	511	19·8	53·5	66·7

* NZ Census 1996, aged ⩾15 years.
† Auckland Regional Public Health Services notification (3 un-geocoded cases excluded).
‡ Auckland Giardiasis Study cases.
§ Standard two-sample capture–recapture equation.

geocoded and mapped and water supply attributes were overlaid. Over 86% of notified cases and 83% of study cases had been drinking water at home from the Auckland Metropolitan Mains (AMM) reticulated water zone, supplying 90% of the regional population. Although, the rates of giardiasis notifications for reticulated and un-reticulated water zones were 52 and 72 per 100 000 population respectively. When data for notified and study cases were overlaid by area of residence and water distribution zones (Fig. 2), a proportion of the study cases did not match with that of notified cases which signifies that these cases had not been notified (Table 3).

When study cases were matched by name, gender, age and address of residence with that of ARPHS notified cases, the sensitivity of case notifications was 49·3%, i.e. 51·7% of study cases were not notified. Analysis of under-notification, as calculated by comparing the total identified cases, found that nearly 20% of cases might not have been notified (Table 3). This calculation was dependent on the proportion of unmatched study cases only.

To estimate under-notification, further analysis was performed by using the capture–recapture method where notified cases were considered to be 'M' and study cases to be 'n' and 'm' for the cases common (matched) between the two samples [28]. The capture–recapture estimation found that the total expected cases should be 829, which was double the number of cases actually notified (Table 4). This estimation suggested that, regionally, 60% of cases of *Giardia* infection in adults ⩾15 years from North Auckland were notified while other regions of Auckland each had 44% notification rates.

This suggests that more than half of the estimated giardiasis cases among the adult population in the ⩾15 years age group from Greater Auckland are likely to have not been notified. Converting this into rates indicates that the true giardiasis infection rate among the adult population in Auckland ought to be 108·3/100 000 population which is two-fold higher than the rate actually notified (53·5/100 000) (Table 4).

DISCUSSION

This study aimed to estimate the proportion of under-notification of giardiasis cases in the Auckland population aged ⩾15 years. Two independent datasets were used to estimate under-notification. Information collected retrospectively as part of a case-control study and as part of routine disease surveillance was compared. The age and gender distributions of people in these datasets were not significantly different. A second peak in the bimodal pattern of giardiasis incidence in New Zealand usually begins after 15 years of age [25] which is preceded by an initial peak in children under 5 years.

The recruitment of study cases was dependent on referrals from GPs. The response rate of study cases was more than 90% of all cases referred by GPs, although, it represented only 50% of notified cases. This may affect the generalizability of the case-control study particularly where characteristics of non-referrals were not known. The requirement to communicate in English and have a contact phone number for subjects may have resulted in some minor selection bias. All cases, except one teenager and one

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Table 4. *Expected number of notified cases by area of residence calculated by capture–recapture methods*

Area of residence	Population* P No.	Notified† M No.	Study‡ n No.	Matched m No. (%)	Cap–recap§ N* No.	Cases/100 000 $M/P \times 10^5$ Current	Cases/100 000 $N/P \times 10^5$ Estimated
Central Auckland	270 195	158	71	31 (43·7)	357	58·5	132·1
North Auckland	160 881	109	62	37 (59·7)	181	67·8	112·5
West Auckland	120 168	42	19	8 (42·1)	95	35·0	79·1
South Auckland	214 542	101	47	22 (46·8)	222	47·1	103·5
Total	765 786	410	199	98 (49·3)	829	53·5	108·3

* NZ census 1996, aged ⩾15 years.
† Auckland Regional Public Health Services notification (3 un-geocoded cases excluded).
‡ Auckland Giardiasis Study cases.
§ Capture–recapture method: $*N = \{(M+1)(n+1)/(m+1)\} - 1$.

non-English speaker, were interviewed successfully irrespective of their ethnicity. Language was not a major barrier for data collection nor the telephone interview as 96% of households in New Zealand have access to a telephone [29]. There was no variation in the recruitment process that could bias the comparability of this dataset with that of notified cases.

Giardia infection shows a seasonal pattern of late summer and early autumn peaks both in New Zealand [25, 30, 31] and overseas [32, 33]. This pattern had a positive influence on the recruitment of cases. However, a reported outbreak of notified cases from a junior school during case recruitment for the case-control study did not noticeably increase the rate of notification of giardiasis [34].

The study quantified giardiasis notified cases in reticulated and unreticulated water supply zones. Given the similar proportion of notified and study cases resident in the AMM area, chances of misclassification between these datasets are small. Water is responsible for a quarter of endemic *Giardia* infections in the United States [35], and the role of contaminated municipal water supplies in epidemic outbreaks of giardiasis is undeniable [36, 37]. *Giardia* cysts are also reported to be abundant in New Zealand waters [38]. However, a recent investigation of the reported outbreak of giardiasis in a Central Auckland primary school did not implicate the mains water supply [34].

As expected, a higher proportion of *Giardia* cases came from Central Auckland, consistent with other data from ARPHS [39]. The rate of infection in Central Auckland was lower compared to North Auckland. The central area is supplied by reticulated water. Nonetheless, the notified infection rate was relatively higher suggesting that factors other than local supply were responsible for the transmission of infection. Given the relatively high proportion of people resident in South Auckland, giardiasis notifications and study cases were comparatively low. The reasons for such low notification are yet to be explored. However, factors such as, a patient's willingness or opportunity to seek medical advice, the GP's decision to request stool samples, a patient's compliance with the request for stool tests, and the GP's compliance with disease notification requirements, would all affect notification rates [35].

It was difficult to ascertain notified cases without having access to laboratory data. Although, diagnostic tests have high sensitivity and specificity, a poor notification rate may still undermine efforts in surveillance and disease control. It is not clear whether complete ascertainment of notified cases from a single surveillance is possible or desirable [40]. Data ascertainment is dependent on the sensitivity of surveillance systems and their attributes such as simplicity, flexibility, acceptability, representativeness and the timeliness of the procedure [41]. Incomplete reporting affects representativeness while the estimation of sensitivity requires exhaustive incidence studies which can be expensive and not useful as a routine disease surveillance tool. The capture–recapture method is a potentially useful alternative to estimate, indirectly, the sensitivity of surveillance data for *Giardia* infection. This study utilised a simple two-source capture–recapture method which highlighted the proportion of non-notified cases. The data from a case-control study and passive surveillance (notification) data bank used in this study were assumed to be independent as they were collected separately from

the same population and for the same period of time. No major shift in population was reported for the year of data collection. The individual cases in this data collection process are thought to have had the same probability of being included in the survey. These assumptions could not be tested directly, and violation of either of them could lead to over- or underestimation of the true notification rate [4]. It is also suggested that truly independent data are hard to find. They usually require a clear understanding of the biology of the disease involved, the dynamics of the reference population, and the assumptions and strengths of the specific methods used [42]. Apart from temporal delay in case notification, other factors influencing the performance of capture–recapture methods are misdiagnoses, over-diagnoses, imperfect and variable 'catchability' and record linkages within sources [26, 43]. All the cases were laboratory diagnosed which reduced chances of misclassification bias. However, the number of cases was higher in the notified group due to the statutory requirement for notification of giardiasis cases by GPs and voluntary referral to the study. We did not match cases by GP clinic but rather used other identifiers such as residence, age and gender. No significant differences were found between the distribution of area of residence, gender or age groups in the two datasets. Three cases from the notification list could not be matched due to lack of details. They were excluded which did not affect the estimation. The sensitivity of 49% for giardiasis notifications as found in the present study was low compared to that for the notification of other infectious diseases using similar estimators [21, 22]. This could be due to disease priority and the perceived importance of diseases by GPs [44]. The magnitude of estimation of cases by capture–recapture method depends on the ascertainment of cases and source dependence. The influence of potential estimation factors, i.e. over- or underestimation could be negligible with high individual ascertainment rates while the presence of negative source dependence may lead to overestimation of the number of cases [26]. Individual case ascertainment is sensitive to completeness of information which directly affects the extent of cases estimation [21]. Since the primary source of data was GPs, they are unlikely to have any significant negative dependence, and furthermore, the individual case ascertainment was high in Auckland irrespective of the completeness of notifications [45]. Therefore, any suspected overestimation in this study is likely to be modest [26].

The number of cases found in this capture–recapture study suggests the real incidence of giardiasis is twice that of cases notified. In other words completeness of giardiasis notifications in Auckland is approximately 50%. Under-notification of giardiasis incident cases has always been suspected [25, 32], but has not yet been estimated. A previous unpublished report by ARPHS which compared a laboratory diagnostic list of *Giardia* positive organisms with notifications found 37% under-notification (Hill P, et al., unpublished report, ARPHS, NZ 2000). Hill's analysis was influenced by a paucity of data from one of the two diagnostic laboratories in Auckland which possibly led to a biased underestimation [46]. Infectious disease surveillance for foodborne diseases in Auckland reported 35% under-notification rates, possibly confounded by outbreaks and the severity of cases [45]. We did not differentiate outbreak cases in this analysis. However, no substantial outbreaks were reported during the data collection period [24].

The present study used the simplest of the capture–recapture methods and found a gross under-notification of giardiasis in the Auckland adult population. This information is useful both for comparing data and for policy makers. Capture–recapture methods are prone to biased estimates if one or a combination of sources captures relatively few cases [46]. Although, this method is not advocated as a replacement for population survey [21, 47, 48] it may be useful in situations where existing databases do not provide complete enumeration of diseases. This is also valuable in evaluating the sensitivity of surveillance systems [21]. Moreover, for international comparison of surveillance data it is helpful to have a knowledge about the sensitivity of the different surveillance systems [49].

It is uncertain whether the application of capture–recapture methods can assess the exact number of cases in the target population [47]. Nevertheless, the estimation of the total number of cases is better with capture–recapture methods than with the traditional register which does not take missed cases into account, presenting only the minimum number of cases [26]. As completeness of disease registration is difficult to achieve, consideration should be given to using a combination of data sources to reduce bias and to achieve the most accurate estimate of incidence [46]. While laboratory-based reporting systems are being recommended to improve completeness of notification, capture–recapture methods can supplement the current process by providing

a cost-effective, accurate and useful adjunct to the disease surveillance system. It is essential to know more accurately the incidence of giardiasis to understand the actual burden of the disease in New Zealand which is already reported to be the highest among developed countries [25, 50]. Such information contributes to the prioritization of work in waterborne and communicable diseases in New Zealand.

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