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Patterns and trends in quality of response rate reporting in case-control studies of cancer

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ABSTRACT

Purpose: We assessed the quality of reporting of response rates in published case-control studies of cancer over the past four decades.

Methods: We reviewed all case-control studies of cancer published in twelve major epidemiology, public health, and general medicine journals in four publication periods (1984-86, 1995, 2005, and 2013). Information on study base ascertainment, data collection methods, population characteristics, response rates, and reasons for non-participation was extracted. Quality of response rate reporting was assessed based on the amount of pertinent information reported, and in particular, numbers of non-participants by reasons for non-participation. We calculated subject response rates by quality of response rate reporting.

Results: A total of 370 studies met the eligibility criteria, yielding a total of 370 case series and 422 control series. Overall, the quality of reporting of response rate and reasons for non-participation was poor. There was a tendency for better quality of reporting of case series, followed by population control series, and lastly by medical source control series. Quality of reporting declined from 1995 to 2013.

Conclusion: The reporting of relevant information on response rates in case-control studies of cancer has been rather poor, and it has not improved over time. This compromises our ability to assess validity of studies' findings.

Key Words: Case-control studies, Cancer, Epidemiologic methods, Response rate, Participation rate

1. INTRODUCTION

In case-control studies, the subject response rate is often used as an indicator of potential selection bias due to non-participation.[1–6] It is believed that response rates have declined over the last decades,[1,3,7,8] particularly among controls.[3,7]

The term "response rate" is defined in different ways, and is often used interchangeably with other terms such as "participation rate" and "cooperation rate".[1] As usually defined in survey research[9] and in epidemiology,[4,10] participation rate is a general term and both "response rate" and "cooperation rate" are particular types of participation rate. The numerator is always the same, the number of subjects who participate, but the denominator can differ. The denominator of the "cooperation rate" should consist of subjects who were contacted, whether or not they agreed to participate; the denominator of the "response rate" should consist of all subjects who were eligible to participate, regardless of whether they could be contacted.[1] This is how we will use the terms. However, there is no agreed upon method to

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define "subject eligibility" in practice; subjects who are unable to be contacted, are in poor health, or for whom contact is not authorised by their physicians, are treated inconsistently by different authors in terms of whether they should or should not be included in the denominator of response rate calculations.[2, 10] Moreover, authors would often not report such information, leaving it impossible for readers to discern fully what was done about different subsets of non-participants.[11, 12] It has been claimed that the quality of response rate reporting is often questionable.[1, 3, 4, 13, 14] The aim of our study was to assess the quality of response rate reporting in published case-control studies of cancer and the evolution of reporting quality over four decades. In practice, our measure of "quality" of reporting is the "amount of pertinent information provided regarding response rates and components of non-participation".

2. Material and methods

2.1 Sample selection

This is a review of questionnaire-based case-control studies of cancer that were published over four decades. In a preliminary exercise we established that PubMed and other automatic search methods were not reliable in identifying all case-control studies, and even less, in identifying those that reported response rates. We realized that we would have to review all articles in certain journals one-by-one. Given the enormous number of studies in all journals and the practical limitation of being able to review them all, we instituted a strategy to restrict numbers but yet maintain relevance. Namely we decided to select a fixed set of journals and a fixed set of calendar years, in order to screen each article in those journal-year combinations. Our team has been active in cancer epidemiology since the late 1970s, and we have amassed a bank of thousands of reprints of journal articles. In order to identify the journals that have been the main vehicles for publication of epidemiological case-control studies of cancer during the period, we conducted an informal survey of our reprints. It turned out that, taken together, the twelve international journals listed in Figure 1 were responsible for a large majority of relevant studies. As these covered a range of journals of epidemiology, public health, and general medicine, we felt confident that these would not have represented a biased sample of possible journals. Some of these journals did not exist for the entire period, reflecting the reality of a shifting pool of journals in which such articles might have been published. We further restricted attention to articles published in certain calendar years in each decade. We aimed to include the middle year in each decade from the 1980s onward. This was possible for 1985, 1995 and 2005. We started this project in 2014; thus we chose 2013 as the approximate "mid-decade" year for the 2010s. Once we began searching for articles, we realized that the number of relevant articles was much lower in 1985 than in the other years. To compensate, we enlarged the 1980s pool to include the three-year span from 1984 to 1986. For those selected journals and those years, we "manually" examined each issue and each article, and selected those that satisfied the following additional inclusion criteria: 1) cancer etiology in adults; 2) conducted in North America, Europe, Australia, or New Zealand; 3) classic case-control design; nested case-control or case-cohort studies were excluded; 4) at least 50 cases or 50 controls, so as to minimize statistical instability of parameters of interest; 5) data collection from subjects or their proxy respondents using questionnaire instruments; pure record linkage studies were excluded; 6) If multiple reports were published in the selected years using the same case and control series, we only included the latest publication in our sample. If the selected publication did not mention subject participation, we sought relevant information from preceding publications by the study team. Figure 1 shows the flowchart of study sample selection. A total of 370 articles satisfied the eligibility criteria.

2.2 Data collection

We reviewed each selected study and extracted the following information: journal name, publication year, data collection period, study location, cancer type, type of control series including population, medical-source (patients from sources such as hospital, clinic, HMO, GP list, cancer or death registers), and friends and/or family control series, mode of data collection (in-person, mail, telephone, or multiple methods), type of respondent accepted (self only, proxy only, or self and proxy), and terminologies used by authors to describe level of subject participation ("response rate", "participation rate", "cooperation rate", or multiple terminologies used). For each case and control series, we extracted the frequency distribution of participation of eligible subjects, including reasons for non-participation. The typical reasons for non-participation are: subject refusal, subject deceased or too ill, subject unreachable, and subject not contacted due to medical source obstacles. Medical source obstacles refer to situations where researchers were precluded from contacting some eligible subjects because either the treating physicians or certain medical/administrative agencies refuse to or are unable to grant access to those subjects. This latter reason for non-participation is generally applicable to cases and to controls selected from medical sources, but not to general population controls.

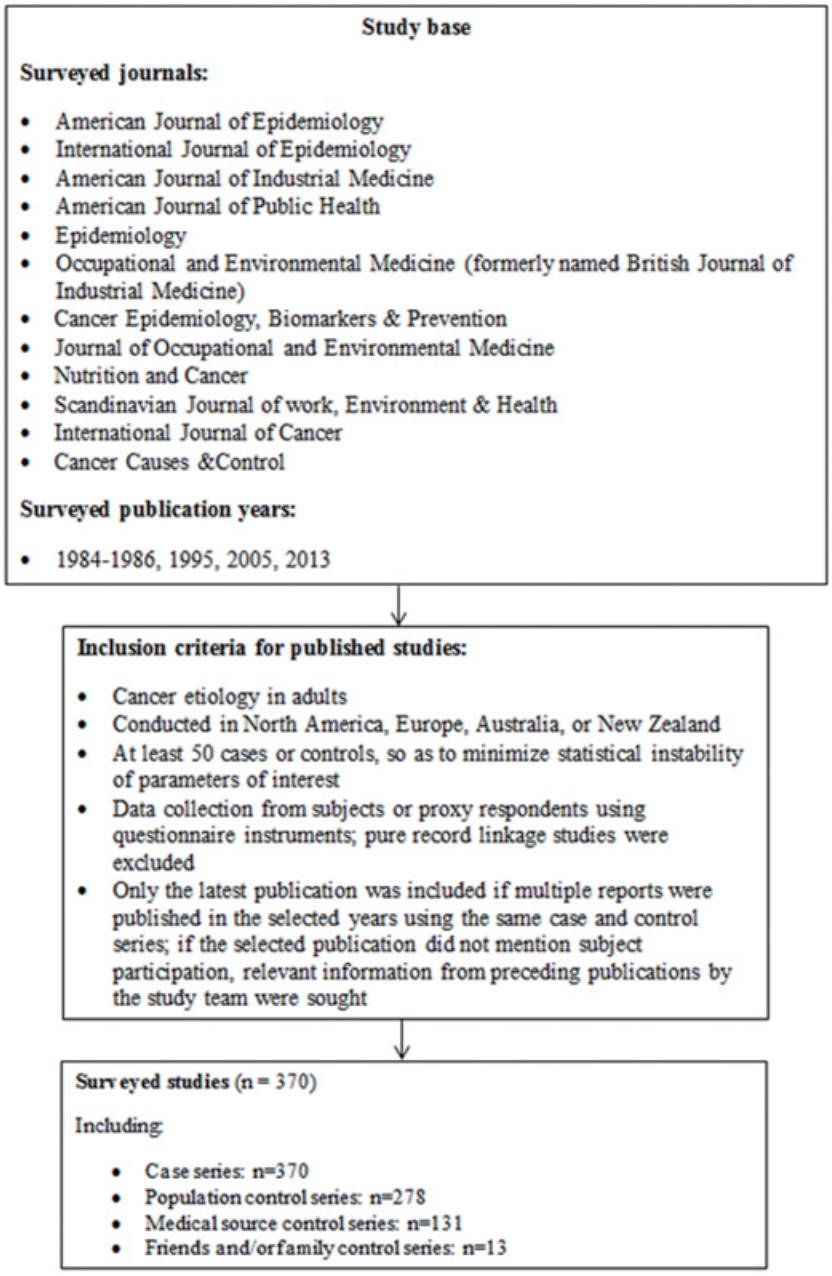

Figure 1. Surveyed study selection method

2.3 Response rate calculation

The response rate can be calculated as follows:

$$Response\ Rate = \frac{Participants}{EligibleSubjects}$$

Failure to include all eligible subjects in the denominator would produce misleading estimates of response rate. The denominator should include all subsets of nonrespondents listed above. Namely:

Eligible Subjects= Participants + subject Refusal + subject Deceased or Too Ill + Subject Unreachable + Medical Source Obstacles (if applicable)

In any given study, one or more of these components of "eligible subjects" might be very small or even null, and this could lead an author to not even mention whether there were any such subjects. However, we believe that a best practice approach to reporting would encourage an author to report whether or not any of these categories existed in the study and how many subjects were in each. Otherwise, it would be difficult for a reader or reviewer to know whether the category is omitted because there were very few instances or because the author chose to exclude such subjects from the list of eligible subjects. In this paper, we will measure the

reporting of response rates against this exacting standard.

2.4 Quality of reporting of response rate

We examined the distributions and time trends in quality of reporting of response rate, separately for each of the following series of subjects: cases, medical source controls, and population controls. The unit of observation was therefore the series of subjects in each publication, with most publications providing two units of observation (a case and a control series) and some publications providing three series (where there were two different control series).

Based on the published information, we created a rating system to evaluate the amount of information reported for each component of eligible subjects; that is, number of respondents, number of subject refusers, number of subjects unreachable, number of subjects deceased or too ill, and number of subjects not contacted because of medical source obstacles. For each series of subjects, we rated the information provided regarding each of these components with one of the following descriptors: "*information provided*" or "*information not provided*". If the information on a given component was not explicitly provided but could be calculated from the information in the paper, we counted that as "*information provided*". Further, for the sum of all the components, the total eligible subjects, we added a possible descriptor "*not clear*". This rating is assigned when authors did not report reasons for non-participation, or when the authors have not made it clear how they dealt with various reasons for non-participation in their response rate calculation.

To facilitate the comparison of quality of reporting between time periods and between types of subject series (*i.e.*, cases, population controls, medical source controls), we assigned an overall score to represent the quality of reporting of response rate in each series of subjects, aggregated over all components. An ordinal score ranging from "0" to "3" represents the amount of pertinent information reported on subject participation in each study. Score "0" indicates that no information was provided on subject participation; "1" indicates that there was information on eligible subjects and participants, but no information was provided on reasons for non-participation; "2" indicates that there was information on eligible subjects, participants, and partial information on reasons for non-participation; and "3" indicates that there was comprehensive information on subject participation, including information on eligible subjects, participants, and all four potential reasons for non-participation: subject refusal, subject deceased or too ill, subject unreachable, and medical source obstacles (when applicable).

We calculated response rates for each type of series (cases, medical sourced controls, population controls) based on our standard formula. We performed descriptive analyses to describe the response rates by quality score category (0, 1, 2 or 3), by type of subject series and by time period. 95% confidence limits on the proportions falling into different score categories were computed by assuming a binomial distribution for each. Spearman rank test was conducted to explore the correlation between response rates and the corresponding response rate reporting quality.

All analyses were performed using SPSS (IBM SPSS Statistics, Version 20.0. Armonk, NY: IBM Corp).

3. RESULTS

As shown in Table 1, one epidemiology journal and three cancer journals accounted for nearly 80% of the 370 studies meeting the inclusion criteria. There were approximately equal numbers of studies selected in the years representing each decade of publication. For statistical stability, we regrouped cancer types by general anatomic and survival characteristics. The most studied cancer types were breast, cervix, and endometrial cancers (22%). Two-thirds of studies were conducted in North American populations, with the rest spread across the other eligible regions. The mid-point year of data collection (not publication year) ranged from 1961 to 2010, with 37% conducted during 1991-2000. The 370 studies provided 370 case series. Fifty one studies used multiple control series, yielding a total of 422 control series in our data, of which 66% were selected from the general population, 31% were selected from medical sources, and 3% were selected from friends and/or family of cases. Results for friends and/or family control series are not presented due to the small number of studies using this approach. Sixty-nine percent of studies collected data from subjects in person, with the rest collecting data through mail, telephone, or multiple methods combined. Only 20% of studies accepted proxy response.

3.1 Quality of reporting of response rates

The reporting of components of response rates is presented in Table 2. For each of these components, we observed the best quality of reporting in cases, followed by population controls, and lastly by medical source controls. In addition, among the case series, only 30% mentioned whether there was a possibility of non-participation because of medical source obstacles, and of those, 79% counted those non-participants as part of the denominator for computing response rates. Among medical source control series, only 10% mentioned whether there was a possibility of non-participation because of medical source obstacles, and of those, 58% counted those

non-participants as part of the denominator for computing response rates (data not shown).

Table 1. Frequency distributions of the surveyed studies

	No.	%
All	370	
Journal		
CEBP	83	22.4
AJE	71	19.2
CCC	68	18.4
IJC	63	17.0
Others *	85	23.0
Publication year		
1984-1986	75	20.3
1995	83	22.4
2005	140	37.8
2013	72	19.5
Cancer type		
Breast, cervix, endometrium	83	22.4
Lung, mesothelioma, respiratory tract	42	11.4
Hematopoietic	36	9.7
Prostate, testicle, penis	32	8.6
Head and neck	30	8.1
Colorectum	28	7.6
Bladder, kidney, urinary tract	27	7.3
Ovary	23	6.2
Stomach, liver, pancreas	22	5.9
Skin	16	4.3
Brain	15	4.1
Others	16	4.3
Study population		
North America (USA and Canada)	245	66.2
Southern Europe [#1]	51	13.8
Northern Europe [#2]	41	11.1
Eastern Europe [#3]	8	2.2
Australia or New Zealand	16	4.3
Multiple	9	2.4
Mid-point year of data collection		
1961-1980	63	17.0
1981-1990	103	27.8
1991-2000	138	37.3
2001-2010	59	15.9
Not mentioned	7	1.9
Type of control series [#4]		
Population[#5]	278	65.9
Medical source [#6]	131	31.0
Friends and/or family	13	3.1
Mode of data collection		
In-person	256	69.2
Mail	36	9.7
Telephone	31	8.4
Multiple methods	43	11.6
Not mentioned	4	1.1
Type of respondent accepted		
Self only	297	80.3
Proxy only	6	1.6
Self and proxy	64	17.3
Not mentioned	3	0.8

* All other journals listed in Figure 1; [#1] Southern Europe: Spain, Portugal, Italy, Greece, France. [#2] Northern Europe: Finland, Sweden, Norway, Denmark, Netherlands, Germany, and United Kingdom; [#3] Eastern Europe: Russia, Poland, Czech Republic, Slovakia, Hungary, Romania, Bulgaria, Turkey, Slovenia; [#4]The sum of the numbers of each type of control series do not add up to 370 because some studies used more than one type of control series; [#5] Includes sources such as population registers, electoral lists, random digit dialing, driver's license, governmental medical insurance lists, and neighbors of cases; [#6] Includes such sources as hospital or clinic patients, HMO or GP lists, and cancer or death registers.

Table 3 shows the frequency distribution of the overall quality score of response rate reporting, according to type of study subject (cases, medical source controls, or population controls) and according to the year of publication (1984-86 to 2013). The case and population control series had a very similar pattern for overall quality of response rate reporting; namely, about 75% of studies were scored as "1" or "2" (mediocre), about 15% were scored "0" (uninformative), and about 10% were scored "3" (excellent). However, medical source control series manifested a different pattern; nearly half of such studies had an overall score of "0", and few of them had scores greater than or equal to "2".

When looking at the evolution of response rate reporting by publication year, there were conflicting messages. The proportion of studies with very poor scores decreased from 1980s to 1990s and remained at that level thereafter. But the proportion of studies with good or excellent scores ("2" and "3" combined) declined steadily from about 40% in the 1980s to about 28% in 2013.

3.2 Subject response rates by quality of reporting

Among cases series, response rates were somewhat lower among studies with good quality of reporting of response rates than among studies with low or medium quality (see Table 4). This was confirmed by a statistically significant ($p < .01$) Spearman rank correlation of -0.21 between reporting quality score and response rates.

4. DISCUSSION

There is variability in the criteria used by different authors to define the denominator for computing response rates. For example, some authors exclude unreachable subjects from the denominator, while others include them.[2] A second example of different approaches to defining the denominator is in the context of studies for which full subject eligibility could not be ascertained at the initial stage, such as in studies using random digit dialing (RDD). Some studies using RDD would try to estimate the denominator as the number of eligible subjects in the screened sample, whereas others would use the number of subjects who are known to be eligible after screening.[5] While the publication dates of the surveyed studies ranged to 2013, the dates of fieldwork of the studies were mainly in the 20th century, and mainly in the era before cellphones became as prominent as they are now. Given the decline in landline telephone use in recent years, RDD is becoming a less attractive strategy for ascertainment of controls. Still, other novel methods such as online survey could engender similar issues to those pertaining to RDD insofar as identifying subject eligibility at the initial stage of contact. A third example is in the situation when cases or controls are

selected from medical sources, and medical staff play some role in authorizing contact with subjects or enlisting their participation. In such a situation there may well be some loss of subjects because the medical personnel were not able or willing to discharge their task. Different investigators adopt different approaches to including or excluding the lost subjects in the denominator.

Table 2. Quality of reporting of information on components of response rates in surveyed studies, by type of subject series (case series, medical source control series, and population control series)

	Cases			Medical source controls			Population controls		
	N* = 370			N* = 131			N* = 278		
	Pertinent information provided for the calculation of true response rate[#1]								
	Yes[#1]	No[#1]	Not clear[#1]	Yes[#1]	No[#1]	Not clear[#1]	Yes[#1]	No[#1]	Not clear[#1]
Component of information	**N (%)**	**N (%)**	**N (%)**	**N (%)**	**N (%)**	**N (%)**	**N (%)**	**N (%)**	**N (%)**
Participants	312 (84.3)	58 (15.7)		71 (54.2)	60 (45.8)		242 (87.1)	36 (12.9)	
Non-participants and reasons for non-participation									
Subject refusal	131 (35.4)	239 (64.6)		29 (22.1)	102 (77.9)		78 (28.1)	200 (71.9)	
Subject deceased or too ill[#2]	108 (29.2)	262 (70.8)		8 (6.1)	123 (93.9)		41 (14.7)	237 (85.3)	
Subject unreachable	91 (24.6)	279 (75.4)		9 (6.9)	122 (93.1)		60 (21.6)	218 (78.4)	
Medical source obstacles[#3]	85 (23.4)	279 (76.6)		4 (3.1)	127 (96.9)		-	-	
Total eligible subjects	44 (11.9)	58 (15.7)	268 (72.4)	1 (0.8)	60 (45.8)	70 (53.4)	30 (10.8)	36 (12.9)	212 (76.3)

* N is the number of series of cases or controls that were examined.

[#1] This represents the amount of pertinent information provided by authors for each component of response rate:

Yes: Information is provided in the paper on this component or that it can be calculated from other provided information.

No: No information is provided in the paper on this component and it cannot be calculated from other provided information.

Not clear: This rating only applies to the identification of total number of eligible subjects. This rating is assigned when authors did not report reasons for non-participation, or when the authors have not made it clear how they dealt with various reasons for non-participation in terms of response rate calculation.

[#2] This could include the subject was deceased or too ill and no proxy was allowed or found.

[#3] This could include the patient's treating physician or certain medical/administrative agencies refusing or failing to grant access to the patient. This component only applies to 364 studies in the case series because 6 studies only interviewed the proxies of deceased subjects.

Table 3. Frequency distribution of overall quality of response rate reporting in surveyed studies by type of subject series (case series, medical source control series, population control series) and by publication year

	Overall quality score (range: 0-3*)								
	0			1		2		3	
	N[#]	%	95% CI	%	95% CI	%	95% CI	%	95% CI
Type of subject series									
Cases	370	15.7	(12.1, 19.8)	40.3	(35.2, 45.5)	32.2	(27.4, 37.2)	11.9	(8.8, 15.6)
Medical source controls	131	45.8	(37.1, 54.7)	32.8	(24.9, 41.6)	20.6	(14.0, 28.6)	0.8	(0.0, 4.2)
Population c ontrols	278	12.9	(9.2, 17.5)	55.8	(49.7, 61.7)	20.5	(15.9, 25.7)	10.8	(7.4, 15.0)
Publication year									
1984-86	154	29.2	(22.2, 37.1)	30.5	(23.4, 38.4)	35.7	(28.2, 43.8)	4.5	(1.8, 9.1)
1995	175	18.3	(12.9, 24.8)	41.1	(33.8, 48.8)	26.9	(20.4, 34.1)	13.7	(9.0, 19.7)
2005	295	17.3	(13.2, 22.1)	48.5	(42.6, 54.3)	23.7	(19.0, 29.0)	10.5	(7.3, 14.6)
2013	155	16.8	(11.3, 23.6)	54.8	(46.7, 62.8)	20.0	(14.0, 27.2)	8.4	(4.5, 13.9)

* The overall score represents the amount of pertinent information reported on subject participation in each study.

[#] N is the number of series of cases or controls that were examined.

The scores are assigned with an ordinal score from 0 to 3 (0 being the least informative).

0: No information on subject participation;

1: Information provided on eligible subjects and participants, but no information on reasons for non-participation;

2: Information provided on eligible subjects, participants, and partial information on reasons for non-participation;

3: Comprehensive information on subject participation, including information on eligible subjects, participants, and all four potential reasons for non-participation: subject refusal, subject deceased or too ill, subject unreachable, and medical source obstacles (when applicable).

Table 4. Response rates in surveyed studies by quality of reporting

	Overall quality score (range: 0-3)												
	Total			1			2			3			
	N*	Median response rate (%)	25-75 percentile	N*	Median response rate (%)	25-75 percentile	N*	Median response rate (%)	25-75 percentile	N*	Median response rate (%)	25-75 percentile	*p*-value for heterogeneity of response rates between scores
Type of Subject series													
Cases	311	77.2	68.0–86.0	148	79.1	71.2–88.0	119	78.4	63.5–86.0	44	73.0	60.5–76.8	< .01
Medical Source Controls	71	86.8	75.0–95.7	43	81.0	73.8–95.0	27	93.0	83.2–96.0	1	63.4	–	.07
Population Controls	241	67.0	54.0–75.5	154	67.0	54.0–76.0	57	67.0	52.9–73.5	30	65.1	49.4–76.4	.79

* N is the number of series of cases or controls that were examined; note that we excluded from this table the series for which no response rate was reported.
Score 1: Information provided on eligible subjects and participants, but no information on reasons for non-participation;
Score 2: Information provided on eligible subjects, participants, and some information on reasons for non-participation;
Score 3: Comprehensive information provided on subject participation, that is, information on eligible subjects, participants, and complete information on non-participants (subject refusal, subject deceased or too ill, subject unreachable, and when applicable, subject not contacted due to medical source obstacles).

Modifying the eligibility criteria for the denominator can lead to very different estimates of the response rate.[15] Further, in the context of case-control studies of cancer where the sampling frame usually differs between case and control series, poorly or incorrectly defined eligibility criteria for cases and controls could cause an unpredictable level of selection bias even when reported response rates are high.

Our review included a large sample of questionnaire-based case-control studies of cancer published in the past four decades, which enabled us to explore the current and past practice of reporting of response rates. We selected for this review 12 journals that in our experience were likely to have published a large fraction of epidemiological case-control studies of cancer. From an initial review, other journals would not have accounted for large numbers of studies of the types we were searching for. Furthermore, the journals we selected probably represent the "best case scenarios" of journals with high quality epidemiology review.

We found rather poor reporting of relevant information on subject response rates in case-control studies of cancer, especially regarding inclusion or exclusion from the denominator of different subgroups of nonrespondents. Previous reviews carried out in the early 2000s reported that about half of the case-control studies provided no information regarding their response rates.[3, 12] Our estimates of the numbers of studies with poor information ranged from about 12% among population control series to 45% among medical source control series. These various reviews are not directly comparable because they not only used author-reported response rates, but they also covered different diseases, different topics, different populations, and different eras. Unlike the other reviews, we also searched for information on study participation from previous methodological publications of our surveyed studies. It also seems that we applied more demanding criteria for judging the presence of excellent reporting.

Comparing reporting quality between the types of subjects, there was a tendency for best reporting of subject participation in case series, followed by population control series, and lastly by medical source control series. In order to understand whether the poorer quality among medical source controls than among cases was a function of the investigators who conducted the different types of studies or of the type of subject series itself, we subdivided the case series into those from studies in which population controls were exclusively used and those from studies in which only medical source controls were used. Comparing these two sub-series of cases, we observed that the quality of the reporting for cases was better in studies that exclusively included population-based controls than in studies that exclusively included medical-source-based controls (see Tables 5 and 6). This phenomenon may be partly explained by the following conjecture. It may be that the practice of reporting of response rates is greater among epidemiologically-trained investigators, as opposed to clinically-trained investigators, and it may also be the case that clinically-trained investigators are more likely to use medical source controls than are epidemiologically-trained investigators. To assess our conjecture, we randomly selected from our sample of 370 studies, 50 studies that used population-based controls and 50 studies that used medical source-based controls, and we recorded (from internet searches) the affiliations and degrees of the first and last authors of each selected study. Although this way of ascertaining the epidemiological expertise that went into the various studies was certainly imperfect, the contrast between the two sets of studies was fair and the results were informative. Of the 50 studies using population controls, the numbers that had both first and last authors with apparent epidemiology training, only one of them with

epidemiology training, and neither first nor last author with epidemiology training were 36, 11 and 3, respectively. Of the 50 studies using medical source controls, the corresponding numbers were 31, 14 and 5, respectively. The quality of reporting was better when the main authors had epidemiology training (10% with highest overall score) than when none of the main authors had epidemiology training (0% with highest overall score). There is thus a slight indication in support of our conjecture.

Table 5. Reporting quality of information on components of response rates of the case series in surveyed studies, presented separately for studies that used medical-source-based controls and population-based controls

	Case series					
	In studies using exclusively medical-source-based controls			In studies using exclusively population-based controls		
	N* = 107			N* = 218		
	Pertinent information provided for the calculation of true response rate[#]					
	Yes[#]	No[#]	Not clear[#]	Yes[#]	No[#]	Not clear[#]
Component of information	N* (%)	N* (%)	N* (%)	N* (%)	N* (%)	N* (%)
Participants	68 (63.6)	39 (36.4)		208 (95.4)	10 (4.6)	
Non-participants and reasons for non-participation						
Subject refusal	27 (25.2)	80 (74.8)		89 (40.8)	129 (59.2)	
Subject deceased or too ill[#1]	6 (5.6)	101 (94.4)		90 (41.3)	128 (58.7)	
Subject unreachable	5 (4.7)	102 (95.3)		73 (33.5)	145 (66.5)	
Medical source obstacles[#2]	3 (3.0)	98 (97.0)		75 (34.4)	143 (65.6)	
Total eligible subjects	0 (0.0)	39 (36.4)	68 (63.6)	41 (18.8)	10 (4.6)	167 (76.6)

* N is the number of series of cases or controls that were examined.

[#]This represents the amount of pertinent information provided by authors for each component of response rate:

Yes: Information is provided in the paper on this component or that it can be calculated from other provided information.

No: No information is provided in the paper on this component and it cannot be calculated from other provided information.

Not clear: This rating only applies to the identification of total number of eligible subjects. This rating is assigned when authors did not report reasons for non-participation, or when the authors have not made it clear how they dealt with various reasons for non-participation in terms of response rate calculation.

[#1]: This could include the subjects that were deceased or too ill and no proxy was allowed or found.

[#2:] This could include the patient's treating physician or certain medical/administrative agencies refusing or failing to grant access to the patient. This component only applies to 101 case series in studies using exclusively medical-source-based controls, because the other 6 studies only interviewed the proxies of deceased subjects.

We observed different practice of reporting for subject eligibility due to medical source obstacles between case and medical source control series. Reports about case series were more likely to acknowledge and properly report obstacles in subject ascertainment due to medical personnel than were reports about medical source controls.

In order to minimize the impact of the varying operational decisions made by authors, we recalculated the response rates based on standardized criteria. Still this was an imperfect procedure, dependent on the information we could glean from the publications. Our observation of slightly lower response rates among cases series from publications with high quality of reporting may well indicate that there was crucial information hidden from view in some publications, in particular those with poor reporting, and that even our revised response rate estimates are too high for those studies.

Although the proportion of studies not reporting any information on response rates has declined slightly over time, the overall quality of reporting, as measured by our scores of "2" or "3", has deteriorated since the 1990s. We conjecture that it improved up to that point because of increasing awareness of the importance of response rate as a contributor to study quality, and it declined afterwards because of increasing challenges to achieving high true response rates, resulting in a greater reluctance to reveal the true response rates to journal editors and reviewers (*i.e.*, the fear that disclosing too much information might harm the prospects for publication), coupled with increasing pressure on word counts. The proportion of studies with excellent reporting remains very low.

Table 6. Overall response rate reporting quality of the case series in surveyed studies by publication year presented separately for studies that used exclusively medical-source-based controls and population-based controls

Case series											
Overall score (Range: 0-3#)		Publication year									
		1984-1986		1995		2005		2013		Overall	
		N*	(%)	N*	(%)	N*	(%)	N*	(%)	N*	(%)
In studies using exclusively medical-source-based controls	0	13	(40.6)	10	(40.0)	11	(28.9)	5	(45.5)	39	(36.8)
	1	11	(34.4)	10	(40.0)	16	(42.1)	2	(18.2)	39	(36.8)
	2	8	(25.0)	5	(20.0)	11	(28.9)	4	(36.4)	28	(26.4)
	3	0	(0.0)	0	(0.0)	0	(0.0)	0	(0.0)	0	(0.0)
	Total	32	(100)	25	(100)	38	(100)	11	(100)	106	(100)
In studies using exclusively population-based controls	0	2	(7.4)	1	(1.9)	3	(3.6)	4	(7.1)	10	(4.6)
	1	6	(22.2)	18	(34.0)	34	(41.0)	33	(58.9)	91	(41.6)
	2	16	(59.3)	20	(37.7)	29	(34.9)	12	(21.4)	77	(35.2)
	3	3	(11.1)	14	(26.4)	17	(20.5)	7	(12.5)	41	(18.7)
	Total	27	(100)	53	(100)	83	(100)	56	(100)	219	(100)

* N is the number of series of cases or controls that were examined.

The overall score represents the amount of pertinent information reported on subject participation in each study.

The scores are assigned with an ordinal score from 0 to 3 (0 being the least informative).

0: No information on subject participation;

1: Information provided on eligible subjects and participants, but no information on reasons for non-participation;

2: Information provided on eligible subjects, participants, and partial information on reasons for non-participation;

3: Comprehensive information on subject participation, including information on eligible subjects, participants, and all four potential reasons for non-participation: subject refusal, subject deceased or too ill, subject unreachable, and medical source obstacles (when applicable)

5. Conclusions

Information on response rates has not been well reported in case-control studies of cancer. It is difficult to properly evaluate the validity of studies, given the lack of transparency and consistency in the reporting and calculation of response rate. Although efforts such as the development of the STROBE statement[14, 16] aim to improve the overall reporting quality of observational epidemiologic studies, the impact of such initiatives on response rates is yet to be manifested.

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Authors' contributions

JS conceived the idea of the study. MX, LR and JS were involved in study design. MX and SC were involved in sample selection. MX performed extraction and interpretation of data, data analysis and drafting of the manuscript. JP provided expert advice in study design and data analysis. MX and JS drafted the submitted manuscript. All authors have read and approved the final version of the manuscript.

Conflicts of Interest Disclosure

The authors declare that they have no competing interests.

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