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Accuracy of case-reported family history of melanoma in Queensland, Australia

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A positive family history is used in clinical practice as an indication of increased melanoma risk, yet there are no data on the accuracy of reported family histories of melanoma. The validity of case-reported family history of melanoma was assessed in the course of a family and twin study of melanoma in Queensland, Australia, conducted among the families of 2,118 melanoma cases diagnosed in Queensland between 1982 and 1990. A total of 913 melanoma cases made 1,267 reports of melanoma among their first-degree relatives. A total of 1,040 of these reports were checked, first through relatives themselves and then, if the relative also said they had had melanoma, through the relative's medical records. Medical confirmation of melanoma as the diagnosis was obtained for 623 reports (59.9%; 95% confidence interval 56.9–62.9): a false-positive reporting rate by cases of 40.1%. The level of false-positive reporting was lower for cases under 70 years of age, for women, for cases whose own diagnosis of melanoma was more than 5 years earlier, and for cases with three or more relatives with melanoma. Media campaigns in Queensland aimed at increasing skin cancer awareness, and confusion between melanoma and other more common actinic neoplasms (basal and squamous cell carcinomas), may partly explain the high false-positive reporting rate observed here. For this reason, it is difficult to generalize these findings to northern hemisphere populations where skin cancer is not such an important public health issue.

Key words: death certificates, family, medical records, melanoma, naevus.

Introduction

Queensland has the highest recorded incidence of melanoma in the world.[1] A public education campaign in Queensland in recent years has aimed to increase awareness of melanoma and to promote the avoidance of sun exposure, the major environmental risk factor for this disease.[2] Despite this, 'melanoma' is a word apparently still poorly understood by the public, and often confused with other types of skin cancer and benign naevi.[3] It has been known for some years that a family history of melanoma is associated with increased melanoma risk.[4–7] This has led to suggestions that patients who present with a positive family history constitute a high-risk group who may benefit from regular skin surveillance aimed at early melanoma detection.[8,9] This assumes that patients are able to report their family histories of melanoma accurately, yet there are no data available to support this. The aim of the present investigation was to evaluate the accuracy of case-reported family histories of melanoma, by comparing cases' reports of melanoma among their first-degree relatives with those relatives' self-reports and medical records.

Materials and methods

Study subjects and data collection

Validation of case-reported family history of melanoma was undertaken during a family and twin study of melanoma in Queensland, Australia: the Queensland Familial Melanoma Project. The design of the family study has been described in detail elsewhere.[10] From the 12,006 first incident cases of cutaneous melanoma, invasive and *in situ*, diagnosed in Queensland residents between 1982 and 1990 and reported to the Queensland Cancer Registry, we were able to obtain doctor's permission to approach 10,407 cases, of whom 7,784 (75%) returned a brief family history questionnaire. A more detailed questionnaire was posted to 2,920 of these, including all who had claimed a positive family history ($n = 1,529$) and an approximate 20% random sample of the remainder ($n = 1,391$; Figure 1). Cases were asked for the names and addresses of all first-degree relatives, relatives' dates of birth and ages, and whether any of these relatives had had 'a melanoma diagnosed by a doctor'. Altogether, 2,118 respondents (73%) belonging to 1,912 separate families named a total of 15,907 family members, including 1,238 with melanoma (Figure 1).

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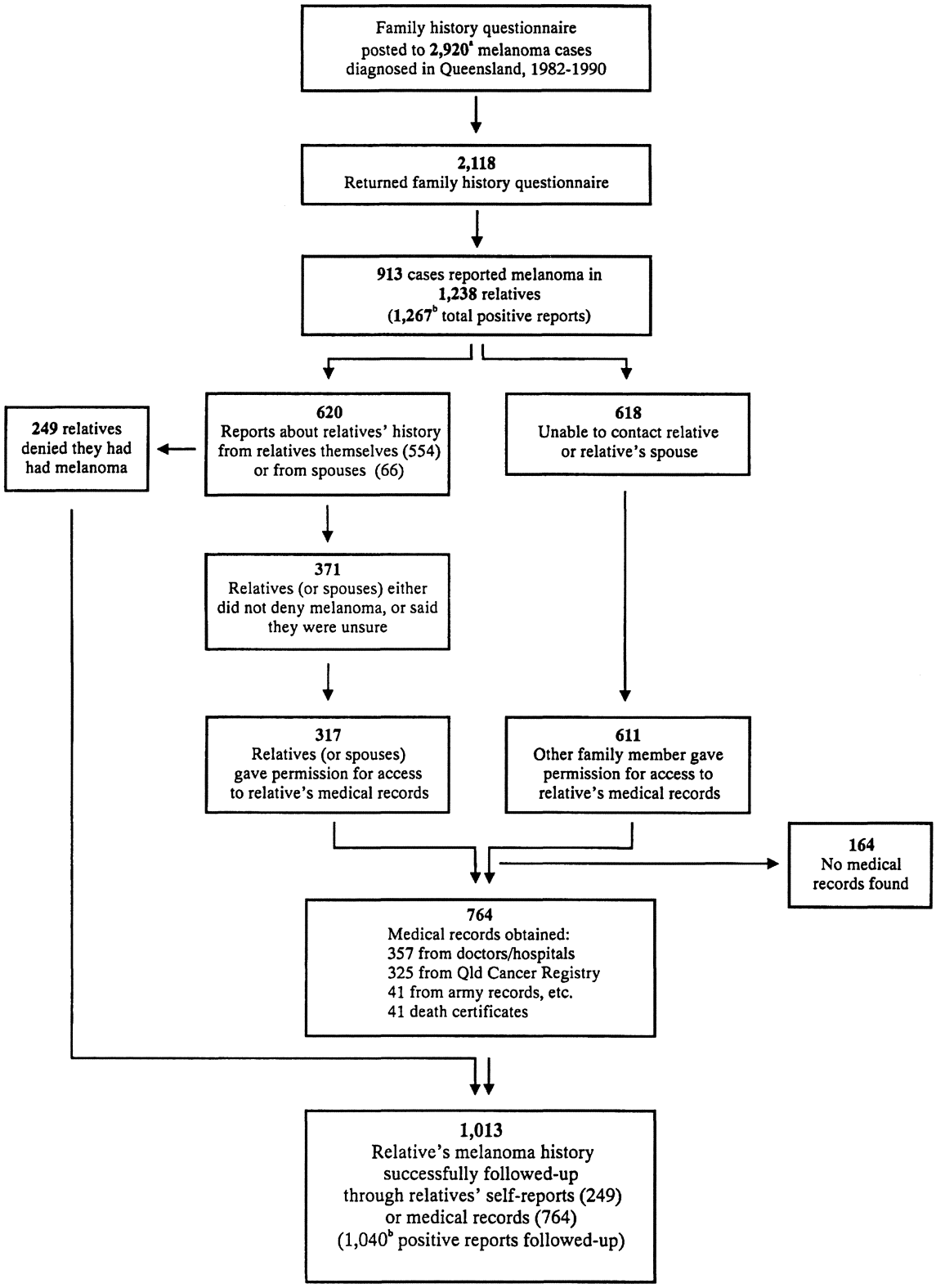

Figure 1. Flow chart illustrating location of relatives for a validation study of reported family histories of melanoma, conducted in the course of a family and twin study of melanoma among 2,118 melanoma cases diagnosed in Queensland, 1982–1990. [a]Includes all cases who reported a positive family history in a brief initial questionnaire ($n = 1,529$) and a 20% random sample of cases who did not report a positive family history ($n = 1,391$). [b]There are more reports than relatives as some related cases reported the same relatives.

A questionnaire was mailed to all 7,619 living relatives between 18 and 75 years of age for whom a respondent had provided a name and address. Of these 5,158 (68%) responded. Relatives were asked whether they had ever been treated for melanoma, for their doctor's name and address, and for signed consent for access to their medical records. Non-responding relatives and the next-of-kin of deceased relatives were contacted by telephone for the same information, including consent to retrieve medical records.

Procedure for confirming reports by cases about their relatives

We sought confirmation for all reports by cases of melanoma in their first-degree relatives as stated in the detailed family history questionnaire. Cases' reports were compared with the relatives' own reports about themselves, and with relatives' medical records. A recorded diagnosis of invasive or *in situ* melanoma was counted as positive confirmation of a case's report. Relatives who stated that they had never been treated for a melanoma were not followed further (Figure 1). Otherwise, after obtaining consent, relatives' doctors were contacted by telephone or letter and asked whether the relative had a history of melanoma. If so, copies of pathology reports were requested. Doctors often named the relative's surgeon or other specialist, or the hospital where the relative was treated and if necessary these sources were contacted in the same way. Records of the Queensland Cancer Registry were checked for relatives' diagnoses. If no information was available from any other source, death certificates were sought for relatives who had died in Queensland.

Data analysis

A confidence interval for the proportion of accurate reports was computed using an exact method.[11] In this calculation, each case–relative pair was treated as an independent set, although a small proportion of relatives were reported by more than one case, and some cases reported more than one relative. Thus, the confidence interval is slightly smaller than would otherwise have been the case.

Results

A total of 1,267 positive reports about relatives were made by 913 cases. These reports concerned 1,238 individual relatives since some related cases reported the same relative. Self-reports by questionnaire or telephone were obtained from 620 of these relatives or their surviving spouses: 249 of these denied a history of melanoma and were not followed further (Figure 1). Permission for access to medical records was obtained for a total of 928 of the remaining relatives, either from the relatives themselves (317) or from other family members (611). Useful medical records were obtained for 764 of these. Sources included the relatives' doctors or hospitals, the Queensland Cancer Registry, army records, interstate cancer registries, pathology laboratories and death certificates (Figure 1). Medical records could not be located for 164 relatives, the most common reasons being that the records had been destroyed under Australian law after a statutory 7-year period, that the relative's doctor had retired, died or could not be located or that the hospital where relatives had been treated had closed.

Thus, in total 1,040 positive reports by 793 melanoma cases concerning 1,013 individual relatives were successfully followed-up, either through the relatives themselves or their spouses (249 relatives) or through medical records (764 relatives).

Accuracy of cases' reports of melanoma in their relatives

Of the 1,040 reports by cases of melanoma in a first-degree relative, medical confirmation of melanoma as the diagnosis was obtained for 623 (59.9%; 95% confidence interval 56.9–62.9). A total of 249 reports were denied by relatives themselves or by their spouses and 168 reports were confirmed in medical records not to be melanoma, giving a total of 417 apparently false-positive reports. Alternative diagnoses obtained from medical records ($n = 148$) or from relatives themselves ($n = 86$) included basal or squamous cell carcinoma (42%), benign naevi (28%), solar keratosis (14%), other cancer (8%), Hutchinson's melanotic freckle (5%) and various other diagnoses (4%), including melanocytic hyperplasia, papilloma and benign keratoacanthoma.

The proportion of positive reports which were accurate increased significantly as the number of years since the case's own diagnosis of melanoma increased ($P < 0.001$), decreased with the age of the case and was higher among women, although neither of these last two trends was statistically significant (Table 1). Cases were slightly more accurate when reporting about their parents and siblings than when reporting about their children. Among cases with a positive family history of melanoma, the accuracy of reports about individual relatives increased as the total number of relatives in the family with melanoma increased ($P = 0.008$). Thus, 83% of reports by cases with a single relative with melanoma were correct, compared with 95% of reports by cases with three or more relatives with melanoma (Table 1).

Table 1. Accuracy of 1,040 positive reports of melanoma in the first-degree relatives of 793 melanoma cases diagnosed in Queensland, Australia, 1982–1990, according to personal characteristics of the cases

Characteristics of cases	Positive reports			*P*-value for trend in proportion of confirmed reports
	By cases about their relatives	Confirmed in relatives' medical reports		
		n	%	
Total	1,040	623	59.9	
Age (years)				
15–30	64	44	68.8	
30–49	376	226	60.1	
50–69	439	262	59.7	
≥70	161	91	56.5	0.170
Sex				
Male	480	273	56.9	
Female	560	350	62.5	0.065
Time since case's diagnosis of melanoma (years)				
<1	91	52	57.1	
1–5	714	401	56.2	
>5	235	170	72.3	<0.001
Type of relative reported				
Parent	338	208	61.5	
Sibling	480	295	61.5	
Child	222	120	54.1	0.106
Number of case's relatives with melanoma				
0[a]	308	0	–	
1	503	419	83.3	
2	134	114	85.1	
≥3	95	90	94.7	0.008

[a]This category was excluded from the test for trend.

Accuracy of relatives' reports of their own melanoma history

Self-reports were obtained from a total of 554 relatives. Of these, 277 reported that they had had melanoma. Medical records were obtained for 275 of these and a diagnosis of melanoma was confirmed for 234 (85.1%). The 41 false-positive reports included 16 basal cell carcinomas, eight squamous cell carcinomas, 13 benign naevi, one Hutchinson's melanotic freckle, one seborrhoeic keratosis and one diagnosis of melanocytic hyperplasia. We could find no recorded diagnosis for one of the false-positive reports. We did not seek medical records when a relative stated that they had not had a diagnosis of melanoma and so are unable to estimate the rate of false-negative reporting by relatives of their own melanoma history.

Discussion

In estimating the accuracy of cases' reports of melanoma among their first-degree relatives we have assumed that relatives who deny a personal history of melanoma are correct. As we did not check negative reports by relatives, and as we are unaware of other studies of the accuracy of negative self-reports of melanoma history, we have no direct evidence to support this, although negative self-reports of other types of cancer have been found to be extremely accurate.[12] Even if 10% of the 249 negative reports by relatives and spouses in this sample were actually incorrect, the estimate of cases' accuracy would only have improved from 59.9 to 62.3%, a small difference. As the true rate of false-negative reporting by relatives is probably much less than 10%, this is unlikely to be an important source of error in this study.

It is more likely that we have over-estimated the accuracy of cases' reports of their relatives' melanoma histories. We attempted to obtain medical confirmation for all melanomas which were reported by cases and not denied by relatives, but were unable to obtain medical records for approximately 20% of such reports. These reports, which were excluded from the analysis, tended to be those concerning relatives who had died or lost contact with the family or whose diagnosis was many years in the past. It would be reasonable to suggest that such reports are probably less accurate than those about living relatives with more recent diagnoses. Thus, our finding that 60% of melanoma reports were correct should probably be regarded as an upper limit for the accuracy of case-reported family melanoma history.

This over-reporting rate of positive family history of melanoma is much higher than that found for reports in first-degree relatives of breast cancer (6%), colon cancer (7 and 22%) and cancer of the pancreas (25%),[12,13] which is somewhat surprising given that melanoma and the result of its treatment, unlike these internal cancers, is usually clearly visible on the surface of the skin. Compared with communities with a lower incidence of melanoma, Queenslanders partly over-report melanoma in their relatives because of their increased risk of other, far more common actinic neoplasms (solar keratoses, basal and squamous cell carcinomas).[14] Confusion between these lesions and melanoma was one of the main sources of error. In addition, given the very high prevalence of melanocytic lesions in this sun-exposed population and the difficulty of distinguishing benign naevi from early melanoma, it is common clinical practice in Queensland to remove melanocytic lesions to exclude melanoma.[15] This, coupled with recent media campaigns in Queensland aimed at increasing skin cancer awareness and the lack of understanding of the term 'melanoma',[3] could have contributed to the high rate of false-positive reporting. It would be difficult to generalize these findings to northern hemisphere populations where skin cancer is not such an important public health issue.

False-positive reporting was more common among respondents over the age of 70 years and among men, perhaps reflecting a better understanding by younger age groups of the term 'melanoma' and a more detailed knowledge among women of the family's medical history. The percentage of false-positive reports also decreased substantially as time since the respondent's own diagnosis of melanoma increased, implying that a recent diagnosis of melanoma (within the past 5 years) is likely to bias a respondent's recollections of their family's melanoma history. Finally, accuracy increased with the number of relatives in the family with melanoma, possibly due to a better knowledge of melanoma among the members of multiple-case families. This comparative over-reporting of melanoma family history among single-case families could result in an over-estimation of the prevalence of familial melanoma in family studies and could potentially bias comparisons between sporadic and familial melanoma cases.

The main implication of these results is that reported family history of melanoma cannot be taken at face value in Australia and should be verified as far as possible. Failure to do so may result in substantial over-estimation of the prevalence of positive family history in epidemiological studies. Clinicians too should bear in mind that reports of melanoma in a close relative have a 40% chance of being incorrect. We have since found that accuracy of reported family melanoma history can be improved by asking the respondent additional questions about how a reported positive relative was treated for their melanoma: in particular was the lesion removed surgically and, if so, how large is the relative's scar. In many instances lesions which are clearly not melanoma can be identified by this means, leaving fewer reports requiring clinical confirmation.

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