
The Western Australian Register of Childhood Multiples: Effects of Questionnaire Design and Follow-up Protocol on Response Rates and Representativeness

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Twin registers have been established worldwide to study the roles of genes and the environment in health and behaviour. While questionnaire surveys are thought to be the most cost-effective way of collecting large amounts of data, low response rates can result in response bias. Many different strategies have been proposed to maximise response rates. A register of all multiple births occurring in Western Australia (WA) from 1980 onwards has been established using probabilistic record linkage techniques. Families who had not experienced the death of one or more of their multiples were invited to participate in the Western Australian Twin Child Health (WATCH) study, which studied the genetic and environmental determinants of childhood asthma and atopy. Several questionnaire designs and follow-up methods were assessed. We have shown that it was feasible to use a population-based register of multiple births to contact families for a questionnaire study. Questionnaire length, mode of follow-up, the number of responses required and the of participants all seemed to affect response.

Studies using twins and higher order multiple births are important for examining the relative contributions that genes and the environment make to a wide range of health-related conditions. This can be achieved either by establishing twin registers or using defined cohorts of twins to study particular outcomes. Many twin registers have now been established world-wide (Boomsma et al., 2002a; Boomsma, 1998). They may collect information on twins of all ages (Skytthe et al., 2002; Stazi et al., 2002), restrict their data to specific age groups (Hayakawa et al., 2002; Kaprio & Koskenvuo, 2002; Spector & MacGregor, 2002; Spinath et al., 2002; Trouton et al., 2002), or be established to study defined populations (Casini et al., 2002; Goldberg et al., 2002; Page, 2002). In

Australia, the Australian Twin Registry (ATR), established in the late 1970s, contains information on over 30,000 twin pairs (Hopper, 2002). It is a volunteer-based register of twins of all ages across all Australian states, and has been used as a sampling frame for research into a wide range of health-related conditions, for example, asthma (Duffy et al., 1990), breast cancer (Boyd et al., 2002), bone density (Hopper et al., 1998), rheumatoid arthritis (Bellamy et al., 1992), dental disorders (Townsend et al., 1998), baldness (Ellis et al., 1998), attention-deficit/hyperactivity disorder (ADHD) (Levy et al., 1997), cardiovascular diseases (Harrap et al., 2000), and melanoma (Zhu et al., 1999). However, the voluntary nature of the register could lead to problems of non-generalizability for some studies, because the expression of many of the conditions of interest to public health research are strongly influenced by factors such as social class, which also modulate response to voluntary data collection (Heath et al., 2001).

Researchers have accessed many data sources when establishing twin registers, including birth records (Anderson et al., 2002; Bergem, 2002; Krueger & Johnson, 2002; Skytthe et al., 2002; Sumathipala et al., 2002), notification of multiple pregnancies from health professionals (Derom et al., 2002; Glinianaia et al., 2002), school records (Anderson et al., 2002), military service records (Goldberg et al., 2002; Page, 2002) and national population or health insurance registers (Bergem, 2002; Kaprio et al., 2002; Stazi et al., 2002; Sung et al., 2002; Yang et al., 2002). Most volunteer-based registers rely on advertising to recruit participants, and this is achieved using various media

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outlets, referrals by health professionals, and contact through twins clubs, parishes, schools and hospitals (Baker et al., 2002; Boomsma et al., 2002b; Hayakawa et al., 2002; Hopper, 2002; Jang et al., 2002; Spector & MacGregor, 2002; Spinath et al., 2002; Strassberg et al., 2002).

Studies in which participants are asked to complete questionnaires at home are more economic than interview studies, but usually have lower response rates (Heath et al., 2001). Cost is an important consideration when a large number of participants need to be recruited and it is therefore very important to maximize response rates. Several strategies have been identified that appear to maximize response to questionnaire surveys (Edwards et al., 2002). These include offering monetary incentives (Collins et al., 2000), using colored ink, sending letters by recorded delivery, using stamps on return envelopes instead of reply-paid (Duffy & Martin, 2001), using a pre-contact alerting potential participants of receiving a questionnaire by mail, making questions “more interesting”, the study having endorsement from a university, and personal follow-up contact (Spry et al., 1989). In recent times, researchers have examined the feasibility of using the Internet to recruit study participants, and assessing any differences in the characteristics of responders (Mavis & Brocato, 1998). However, data about which methods result in the highest response rates are inconsistent. The decision on which methods to use are usually situation-specific and are largely dependent on the nature of the study being undertaken (Larroque et al., 1999), and factors such as cost, timeliness and convenience of data collection and study power (Morris et al., 2001). It is also important to be able to compare the characteristics of responders and non-responders so that any results can be generalised to the wider population. Several factors have consistently been shown to affect response. These include age, gender, educational levels, employment status and urban residence (Heath et al., 2001; Lamers, 1997; Larroque et al., 1999), but the nature of the originating institution, the use of personalised letters and providing feedback to participants also seem to be important (Morris et al., 2001).

A systematic review of published papers examined a number of methods thought to influence response to mailed questionnaires (Edwards et al., 2002). However, of the 292 trials reviewed, only 32% were from medical, epidemiological or health-related journals. Of these, it was not clear how many related to contacting doctors and other health professionals or members of the general population regarding their experiences. Age of study participants was also not known. The combination of various strategies was not considered.

In this paper, we describe the methods we used to trace and contact families belonging to a population-based register of twins and higher order multiples born in Western Australia (WA) between 1980 and

1995. We then assess whether different questionnaire designs and follow-up protocols had an effect on response rates; and determine which factors, if any, influenced response.

Methods

Establishing the WA Twin Register

The Maternal and Child Health Research Database (MCHRDB) is maintained by the Telethon Institute for Child Health Research and comprises records of all births in WA from 1980 onwards (Stanley et al., 1994). Data are derived from the Birth Registration records and the Midwives' Notification of Birth record, and combined to form a composite record for each birth. The MCHRDB is updated when data become available. Records are then linked to the WA registration of deaths and hospital discharge records. As well as prenatal information about the mother during the index pregnancy, the MCHRDB contains data on labor and birth, some postnatal complications and all hospital admissions during childhood.

Multiples born in WA from 1980 onwards were identified using a code for plurality, and verified using a computerized record linkage of sibships of the MCHRDB (Croft et al., 2002). Initially, the WA Twin Register consisted of all multiple births in WA from 1980 to 1992 inclusive, using a grant from the WA Health Promotion Foundation (Healthway). The main purpose for establishing the Register was to invite families to participate in the WA Twin Child Health (WATCH) study which examined the roles that genes and the environment play in the link between childhood asthma and atopy, and exposure to environmental tobacco smoke. The Register has since been extended to include 1993–1995 births, using part of a grant from the Australian National Health and Medical Research Council (NHMRC) for the “WATCH for Asthma” (WFA) study. This study aimed to collect detailed clinical asthma phenotype data on twins born between 1990 and 1995, and their families, and to investigate and describe the familial aggregation of childhood asthma and atopy. We aimed for approximately 60% response to give sufficient power. The different questionnaire designs and follow-up methods adopted for the two studies allowed us to examine the effect they had on response rates and response bias.

Questionnaire Design

1980–1992 Births

Three separate questionnaires were developed: one for the multiples, another for their parents, and a third for siblings of the multiples. Each questionnaire was lengthy (over 100 questions on each), and designed to collect detailed data on asthma and allergy phenotypes in multiple-birth families (the focus of the WATCH study), as well as being used as screening tools for possible future studies of health conditions other than asthma and atopy. They also

contained questions about known risk factors for asthma and allergies, especially active smoking and exposure to passive smoking, as well as a number of questions on general health conditions (including ADHD, epilepsy, physical disabilities, birth defects); occurrence of accidents; use of medications, and use of health-related services. The children's questionnaires (for both the multiples and their siblings) contained additional questions relating to each pregnancy (including the use of assisted reproduction technology, complications of pregnancy, mother's smoking during pregnancy), birth (including mode of delivery, complications and treatment immediately after birth, birthweight, gestation, maternal postnatal depression), duration of breast feeding and age at introduction of other milk products; current weight and height. We also asked a series of questions dealing with the children's education, and parental satisfaction with their children's educational experiences. The multiples' questionnaire also contained a series of standard questions used to determine zygosity (Cohen et al., 1975). Questionnaires for the parents included additional questions on demographics (including education and employment); family history of multiples; household rules affecting family members' exposure to passive smoking; and the reproductive history of the mother. Parents were also asked to complete a table that allowed us to determine the family structure. In total, the questionnaire for the multiples contained 131 questions each, the parents, 107 questions each, and the siblings, 118 questions each. Families estimated that it took them up to 2 hours to complete their series of questionnaires.

Some families withdrew from the study after receiving the questionnaires because they felt they were too long, and they could not spare the time to complete them. In July 1998, therefore, a shorter version of the questionnaire was developed: one questionnaire per family, containing a total of 65 questions. Questions were included if they directly related to ascertaining the prevalence of asthma and atopy, exposure to active and passive smoking, or were needed to determine the family structure. This shortened questionnaire covered all family members and took an estimated 30 minutes to complete. Families contacted at follow-up were offered this shorter questionnaire as an alternative to the set of long questionnaires.

1993–1995 Births

One questionnaire was developed to cover all members of the family and was considerably shorter than those used for the 1980–1992 cohort. As it was used as a screening tool for the WFA study, the questions mainly related to the development of asthma and allergies, along with known risk factors (birthweight, gestation, passive and active smoking, duration of breast-feeding, introduction of other milk, etc.). Details of the family structure were also collected. The questionnaire contained a total of 39 questions and took an estimated 15 minutes to complete.

Tracing and Follow-up Procedures

All Multiple Births

Permission to use WA Department of Health data was given by the Confidentiality of Health Information Committee (CHIC). The study complied with the national Privacy Principles, and was approved by the relevant Institutional Ethics Committee. It was a requirement of CHIC that families who had experienced the death of one or more of their multiples were not to be contacted. A separate study looking at grief and loss in multiple birth families has been conducted (Swanson et al., 2002). Identifiers of eligible mothers (full name, including previous surnames, date of birth, and address at the time of the index birth) from the MCHRDB were linked to the WA Electoral Roll to determine current addresses.

1980–1992 Births

We began the tracing of and mailing to families in 1997. Each mother was sent a letter and information sheet explaining the WATCH study, and asked to indicate her willingness to participate in the study by returning an "Expression of Interest" form in a pre-paid envelope. On this form, she was given the option of providing the name and address of the multiples' biological father if he no longer lived with them. Fathers so identified were then contacted separately. After one month, if there was no response from the mother to this first letter, the White Pages were searched, and if a telephone number was found, the families were contacted by telephone. If not, a follow-up letter was sent to the same address. One further attempt to contact any family who had not replied was made after another month, either by telephone or letter, after which time any family still not responding was considered a non-participant.

Families who agreed to participate in the study were then sent their questionnaires. If they had not returned them after 2 months, they were contacted by telephone (the preferred method) or by letter, and then at monthly intervals until either the questionnaires were returned, the family decided to withdraw from the study or it became impractical to continue follow up. Extra copies of the questionnaires were sent to those families who had misplaced the originals. It was decided to discontinue follow-up on families who did not return questionnaires despite receiving at least three sets of questionnaires, and after several attempts to contact them had failed.

Because more families could be contacted by telephone in the evenings than during the day, follow-up telephone calls were usually made between 6 p.m. and 8 p.m. Monday to Thursday inclusive. In addition, annual newsletters were sent to all families who had agreed to participate, and, in an attempt to elicit their response, to those families who had failed to respond.

1993–1995 Births

Tracing commenced in 2000 and is continuing. Families whose index birth record could be successfully linked to the Electoral Roll were contacted. They

were mailed an introductory letter and information leaflet explaining the study, together with a short questionnaire to complete at home and return in a pre-paid envelope. The mother was also asked to complete and return a registration form, confirming her informed consent to take part in the study. As before, she was also given the option of providing the contact details of the multiples' biological father if he no longer resided with them. Any father so identified was contacted separately. After 1 month, the names of non-responders were searched for in the White Pages. Families were contacted either by telephone or sent another questionnaire to complete. One more attempt to contact non-responders was made, after which time the family was considered a non-participant. Those contacted by telephone were given the option to complete the questionnaire by phone, which took approximately 15 minutes, return the questionnaire by mail, or receive another copy. Families who requested that another questionnaire be sent to them, were further contacted on a monthly basis until the questionnaire had been returned, or the family had decided to withdraw from the study.

Results

A total of 9640 multiple-birth children, born in WA between 1980 and 1995 inclusive, were identified, representing 2.5% of all births during that time. They comprised 4610 sets of twins, 138 sets of triplets, quadruplets and quintuplets (Table 1). Twenty-five families had two sets of multiples during the time period. The percentage of higher order multiples (triplets plus) was about 3% of all multiple births, ranging from a low in 1980, and peaking at 6.1% in 1989.

Of the 9640 multiple birth children, 650 (7%) of them were known to have died. Five hundred and sixty-nine were either stillborn or died within the first 4 weeks of life, giving a perinatal death rate of 59.0

Table 1
Number and Plurality of Multiple Births by Year of Birth

Year of birth	Total	Sets of Twins	Sets of Triplets+*
1980	204	203	< 5*
1981	239	236	< 5*
1982	235	231	< 5*
1983	241	236	5
1984	253	246	7
1985	275	266	8
1986	277	268	9
1987	303	295	8
1988	342	326	16
1989	361	339	22
1990	312	308	< 5*
1991	324	318	6
1992	342	330	12
1993	349	335	14
1994	343	333	10
1995	349	340	9
Total	4748	4610	138

Note: *exact numbers not given to protect the privacy of families

per 1000 births (Table 2), which ranged from a low of 42.3 per 1000 births in 1995, to 77.3 per 1000 births in 1985. Higher order multiples (triplets, quadruplets and quintuplets) had a higher rate of both total childhood deaths and perinatal deaths when compared with twins (10.5% vs. 6.6% ($p = .002$) for all deaths, and 10.0% vs. 5.7% ($p < .0001$) for perinatal deaths, respectively).

The average age of mothers at the time of the multiples' birth was 28.6 years (range 15–44 years). Mothers who were under 20 years of age at the time of the multiples' birth were more likely to have experienced the loss of one of their multiples compared

Table 2
Deaths Among Multiple Birth Children

Plurality	Children	Total deaths	Death rate per 1000 births	Perinatal deaths (PND)	PND rate per 1000 births
Twins	9220	606	65.7	527	57.2
Triplets+	420	44	104.8	42	100.0
Total	9640	650	67.4	569	59.0

Table 3
Deaths of Multiple Birth Children by Mother's Age at Birth of Multiples

Mother's age (years)	Total births	Deaths	Percentage	Perinatal deaths	Percentage
Under 20	434	59	13.6	47	10.8
20–29	5476	364	6.6	331	6.0
30–39	3633	219	6.0	186	5.1
40+	97	8	8.2	5	5.2
Total	9640	650	6.7	569	5.9

Table 4

Number of Families Responding to the Introductory Letter — 1980–1992 Births

	Participant	Non participant	No response	Total
Initial letter only	1067	119		1186
Follow-up by phone	793	82		875
Follow-up by letter	255	79	646	980
Total	2115	280	646	3041

Table 5

Number of Families Returning Completed Questionnaires — 1980–1992 Births

	Completed	Withdrawn	No response	Total
One set only	836	18		854
One set + phone call	494	103		597
One set + f/up letter	21	2		23
Two sets – long, long	298	41		339
Two sets – long, short	163	38		201
Two sets – short, short	15	7		22
Multiple contact			79	79
Total	1827	209	79	2115

with other mothers (13.6% vs. 6.4% ($p < .0001$) for all childhood deaths; 10.8% vs. 5.7% ($p < .0001$) for perinatal deaths) (Table 3).

We were able to trace over 90% of families and invite them to join the WATCH study. Overall, completed questionnaires have been received from 57% of families. A greater proportion of families whose multiples were born between 1993 and 1995 returned completed questionnaires compared with those born between 1980 and 1992 (62% vs. 55%, $p < .001$). This is despite a smaller proportion of eligible families of the younger cohort having been contacted (92% vs. 87%, $p < .0001$) (Figure 1).

Of the 3041 eligible families of multiples born between 1980 and 1992 to be contacted, only 1186 (39%) of them responded to the initial letter without further prompting and 90% of them agreed to participate (Figure 1). Of the remaining families, 875 were contacted by telephone and the remaining 980, by letter, with 91% and 26% respectively, participating (Table 4). Telephone calls made during the daytime were not answered, or resulted in us leaving messages inviting families to return our call. As only a small number of families did so, we felt that daytime calls were not the most efficient use of our limited staff and funds, and we made the calls during the evening wherever possible. Six hundred and forty-six families (21%) did not reply after four attempts at contact. This resulted in an effective participation rate of 70% of the 3041 families who were contacted (Table 4). Questionnaires were sent to the 2115 families who agreed to participate, and 86% of them returned completed questionnaires, representing 55% of all eligible

families (Table 5). Only 40% (836) of families returned their questionnaires without further prompting.

A total of 491 families (27%) responded to the initial letter and returned their questionnaires with no follow up required at either stage. Six hundred and forty-one families (30%) were sent more than one set of questionnaires. In total, 79 families failed to return their questionnaires after several contacts from us, and having been sent questionnaires on three separate occasions (Table 5). It was decided to discontinue follow-up on these families. Eighty seven percent of families who returned completed questionnaires indicated they were willing to be contacted about other studies in the future (Figure 1).

In the 1993–1995 cohort of multiples, only 335 (35%) of the 971 eligible families contacted responded to the initial letter without follow up (Figure 2), with 310 (93%) of them agreeing to participate (Table 6). Most of the remaining families were sent another questionnaire by mail and 65 families completed their questionnaires at the time of the telephone follow-up (Table 6). Ninety-eight families (12%) had not replied after three attempted contacts. A further 25 families have not yet responded and follow-up is continuing. Completed questionnaires have been received from 607 (96%) of the 631 families who agreed to be part of the study (Table 7) and 585 (96%) stated that they were willing to be contacted in the future (Figure 2).

The two-stage contact process used for the 1980–1992 cohort of families inherently meant that this group had a longer time between initial contact from us and our receipt of completed questionnaires. However, families of multiples born between 1980 and

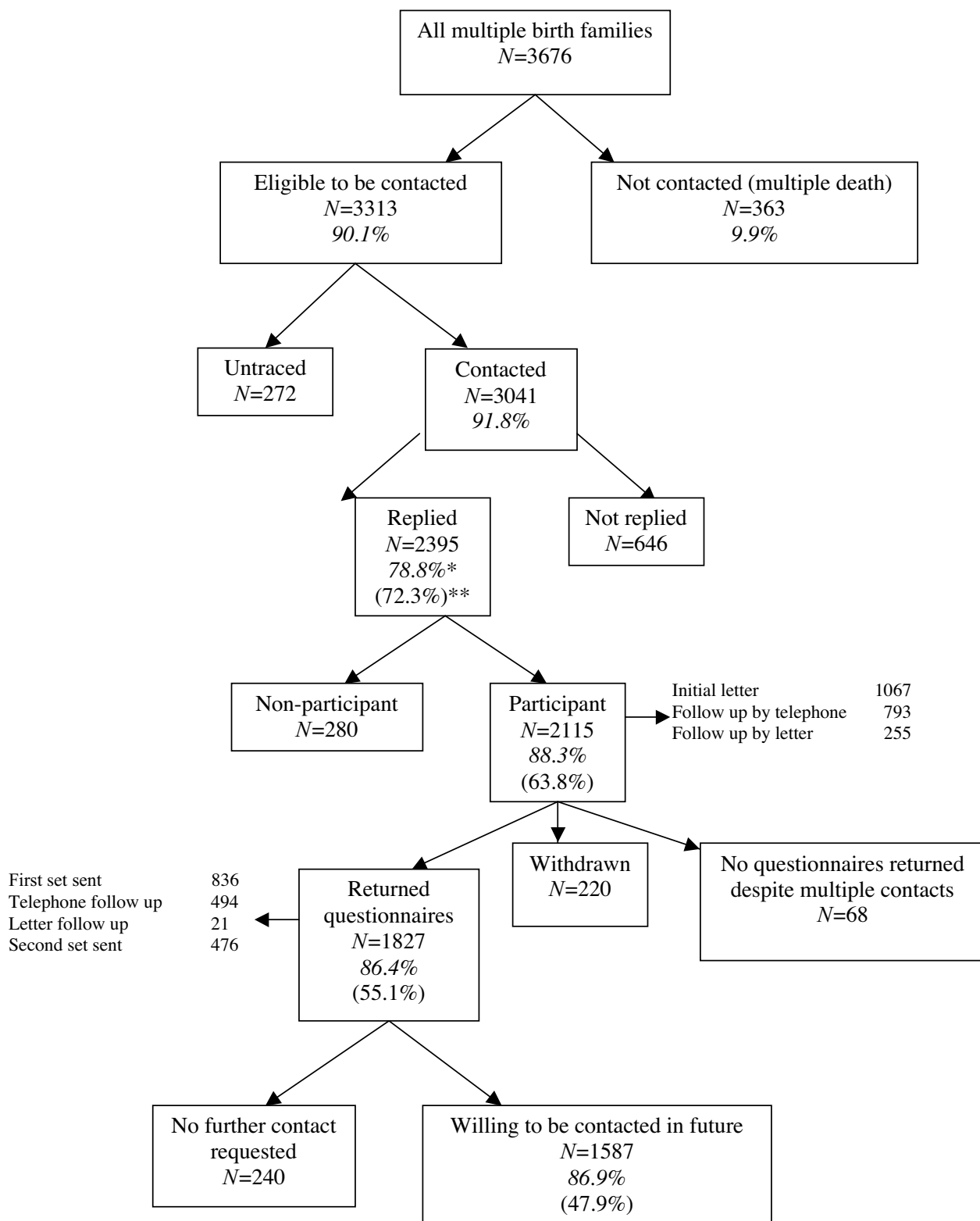


Figure 1
The WA Twin Register, 1980–1992.

Note: * percentage of level above
** percentage of number eligible

Table 6

Number of Families Responding to an Initial Letter — 1993–1995 Births

	Participant	Non-participant	Withdrawn	No response	Total
Letter only	310	25		6	341
Phone follow-up	59	6			65
Letter follow-up	182	41		19	242
2 follow-up contacts	80	17	1	98	196
Total	631	89	1	123	844

Table 7

Number of Families Returning Completed Questionnaires — 1993–1995 Births

	Completed	Withdrawn	No response	Total
One questionnaire only	289			289
Phone follow-up	47	1		48
Two questionnaires	271	2	21	294
Total	607	3	21	631

1992 took more than three times longer to return questionnaires than families of multiples born between 1993 and 1995 (217 days vs. 60 days) (Table 8). Nearly half of all families whose multiples were born between 1993 and 1995 took less than four weeks to return completed questionnaires from the time they received the initial letter. In contrast, more than half of the 1980–1992 multiple-birth families took in excess of 6 months to return their questionnaires.

When considering the length of time between sending questionnaires to families and receiving them

back, families of multiples of the older cohort took slightly longer to return completed questionnaires than families of multiples born between 1993 and 1995 (78 days compared with 60 days) (Table 9). However, the percentage of the older cohort who still took longer than 6 months was significantly greater when compared with the younger cohort (11% vs. 8%, $p = .03$).

In total, 2286 families (57%) had changed their address between the time of their multiples' birth and our contact with them. Families of the younger multiples were less likely to have changed their address (50% vs. 59%, $p < .0001$). Mothers who were under 20 at the time of the multiples' birth were less likely to reply to the introductory letter, and less likely to agree to participate than older mothers (45.2% vs. 79.1%, $p < .001$; 41.7% vs. 70.4%, $p < .001$). They were also less likely to return completed questionnaires (29.8% vs. 61.0%, $p < .001$) (Table 10).

Compared with families living in rural areas of WA, families who lived in the Perth metropolitan area were more likely to respond to the initial letter (79.9% vs. 75.5%, $p = .007$), but no more likely to participate in the study (69.6% vs. 69.0%, $p = .76$). Families who lived outside of Perth were more likely to return completed questionnaires (89.9% vs. 84.6%, $p = .002$) than those who lived in Perth. There was no difference in the overall response rates between metropolitan and rural families (58.9% vs. 62.0%, $p = .11$) (Table 11).

Table 8

Length of Time Between Initial Contact and Return of Completed Questionnaires

Time	1980–1992 births	1993–1995 births
< 2 weeks	39 (2%)	155 (26%)
2–4 weeks	136 (8%)	129 (21%)
4–13 weeks	374 (21%)	190 (31%)
13–26 weeks	306 (17%)	85 (14%)
Over 26 weeks	944 (52%)	47 (8%)
Average	217 days (31 weeks)	60 days (9 weeks)

Table 9

Length of Time Between Sending and Receiving Questionnaires

Time	1980–1992 births	1993–1995 births
< 2 weeks	376 (21%)	155 (26%)
2–4 weeks	323 (18%)	129 (21%)
4–13 weeks	604 (33%)	190 (31%)
13–26 weeks	327 (18%)	85 (14%)
Over 26 weeks	199 (11%)	47 (8%)
Average	78 days (11 weeks)	60 days (9 weeks)

Discussion

It has been shown that it is feasible to establish a population-based register of multiple births in WA, using probabilistic record linkage techniques (Croft et al., 2002). Here, we have demonstrated that it is possible to use this register to contact families for a

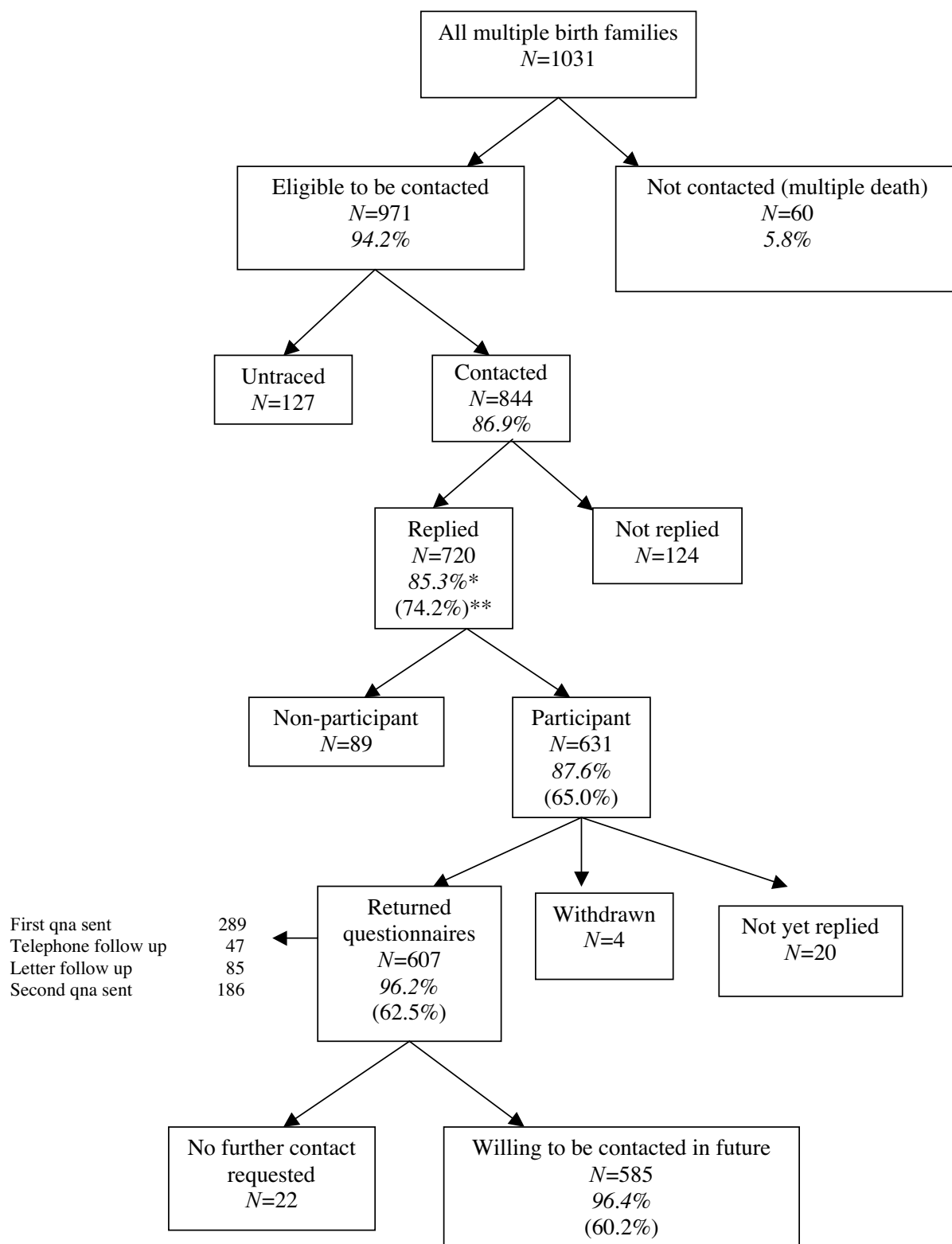


Figure 2
The WA Twin Register, 1993-1995.

Note: * percentage of level above
** percentage of number eligible

questionnaire survey, achieve acceptable response rates, and that respondents appear to be representative of all WA multiple birth families.

The MCHRDB, established and maintained by the Telethon Institute for Child Health Research, is a very powerful resource and consists of data on all children born in WA since 1980 (Stanley et al., 1994). Its unique nature has enabled us to compare participants and non-participants and to examine the representativeness of the register. We have shown that there is no difference between respondents and non-respondents with respect to maternal age at first birth, paternal age at first birth, and racial origin (Hansen et al., 2000). Our overall response rate of 57% is lower than that usually considered acceptable for such studies (Howell et al., 2003), but was adequate in terms of study power.

Tracing people, especially women, can be difficult when the population is mobile and name changes not infrequent. Our attempts to trace and contact families commenced in 1997 when the multiples were aged between 5 and 17 years. The names and date of birth of the mother were used to link to the WA Electoral Roll and subsequently to search the White Pages. However, both sources of data have limitations. To be registered on an Electoral Roll, people must either be an Australian citizen, or be British subjects enrolled before 25th January 1984. The WA Electoral Roll covers approximately 86% of the Western Australian population.¹ It contains information on full name, date of birth and current address. At the time the 1980–1992 birth records were linked, the year of birth was rounded to the closest year ending in either “0” or “5”. The White Pages is of limited use for tracing females, as many women do not have telephone numbers registered in their own name. We also used the Commonwealth Electoral Roll to search for women resident in states other than WA. This is available for public perusal at State Electoral Roll Offices, but as it has no date of birth listed, is of limited use. The Commonwealth Electoral Rolls and White Pages

are most useful for tracing residents who have unusual surnames. We believe that most of the mothers that we have been unable to trace have moved away from WA. However, we still managed to find a current address for over 90% of families, a large number of whom had changed their residential address since the birth of their multiples.

When assessing factors influencing response, this study has shown that:

- there was a lower response to the series of three questionnaires per family, than to the shorter questionnaire that covered all family members
- response was increased if families only had to reply to one mailing, that is, they received their questionnaires along with information leaflets and consent forms, rather than having to respond twice, firstly by returning an expression of interest form and then, returning their completed questionnaires
- telephone was the preferred mode of contacting non-responders
- telephoning families at home was most productive when it was conducted during the early evening hours
- giving potential responders the option of completing their questionnaires by telephone was well received by families. This option appeared only to be feasible for the shorter version of our questionnaires.

What is still unclear, however, is which of these factors had the most impact, and whether they operated singly or jointly. We used a combination of strategies, and were unable to examine the effect of any of them in isolation from the others.

The length of the series of questionnaires sent to the families of the 1980–1992 birth cohort did affect the response rate. Families were happy to agree to participate in the study, but some decided to withdraw after receiving the questionnaires, when they realised the amount of time needed to complete them. The shorter questionnaire was readily accepted and

Table 10

Response Rates by Mother's Age at Birth of Multiples

Mother's age	Contacted	Replied	Agreed to participate	Returned questionnaires	Overall response rate
Under 20	84	38 (45%)	35 (42%)	25 (71%)	30%
20–29	1660	1278 (77%)	1128 (68%)	990 (81%)	59%
30–39	1244	1036 (83%)	915 (74%)	781 (85%)	63%
40+	49	41 (84%)	36 (74%)	31 (86%)	63%

Table 11

Response Rates by Place of Residence

Place of residence	Contacted	Replied	Agreed to participate	Questionnaires returned	Overall response rate
Metro	2102	1680 (80%)	1463 (70%)	1238 (85%)	59%
Rural	885	668 (76%)	611 (69%)	549 (90%)	62%

resulted in a lower percentage of families subsequently deciding to withdraw. Other studies have found that shorter questionnaires result in higher response rates (Bean & Roszkowski, 1995; Edwards et al., 2002), although there seems to be no agreement as to the optimal questionnaire length. We decided to persevere with the longer version of the questionnaires as a first resort, as we felt that the data obtained would be very useful in identifying families for future studies into other health outcomes.

As with some other studies (Mueller et al., 1986; Parker et al., 2000), we found that the most efficient method of follow up contact was by telephone. Many families appreciated the personal contact, and commented that they were pleased that researchers were taking an interest in "ordinary families like ours". However, the telephone calls needed to be made in the early evening hours when most families could be contacted.

The use of incentives to increase response is a contentious one. Many surveys use incentives to motivate volunteers (Collins et al., 2000; Spry et al., 1989), although others who offer incentives target specific groups such as physicians (Tambor et al., 1993; Ward et al., 1998). One study of twins offered monetary rewards to registrants (Jang et al., 2002). However, researchers should take into account the likely impact on the total cost per completed response, and whether the respondents are representative of the population being sampled. It could perhaps even be argued that a university-based study offering a monetary incentive would be seen by potential participants as unethical, or a waste of taxpayer's money, or both. This could actually deter people from participating. Hence, we did not consider using monetary rewards or other incentives. This study originated from the Telethon Institute for Child Health Research, a non-government research institute affiliated with the University of WA, and was funded solely by nationally-competitive grants.

One of the most important aspects to consider when undertaking a large questionnaire study is the characteristics of the study staff. We found that some important requirements for staff carrying out follow up included having a special telephone manner; the ability to be able to devote 2–3 hours during the evening to contact families; empathy/sympathy with the needs and concerns of multiple-birth families; and being a willing listener. Of course, they must strictly adhere to the study follow up protocols, and pay particular attention to detail when using the Electoral Rolls and White Pages to trace mothers. The nature of telephone contact should be friendly, especially if non-responders are to be contacted regularly. Many families require time to decide whether or not to participate in the study. Multiple-birth families have many demands made on their time, but we have found that many mothers have appreciated the opportunity to be able to discuss some of their concerns with a staff member.

This study has also shown that families of multiples born in WA between 1993 and 1995 are more likely to join the WA Twin Register and more likely to return completed questionnaires, than families of WA multiples born between 1980 and 1992. Several factors could account for this. For the younger cohort, questionnaires were mailed with an introductory letter and information package, which meant that families only had to respond to one letter. Also, questionnaires were significantly shorter, and therefore took much less time to complete. At the time the families were contacted, mothers were younger and their multiples were aged between 7 and 8; details of the pregnancy and birth were more recent and therefore more likely to be remembered. Several other studies have also found that the age of respondents effects response, (Larroque et al., 1999) as do the number of mailings (Sauerland & Neugebauer, 2002).

The reasons why families living outside the Perth metropolitan area were more likely to respond than their non-metropolitan counterparts are unclear. Multiple-birth children are more likely to be born prematurely, and at a lower birthweight, than singleton children, and are therefore of special interest to researchers looking at the long-term sequelae of adverse birth outcomes. They are often invited to participate in studies that require them to attend clinics in Perth. But limited research funds do not allow for any reimbursement of costs incurred, and so families living at great distances from Perth, often over 1000 kilometres away, are usually unable to participate. It is possible that metropolitan families could feel that they already take part in enough research, and this could explain why, in this case, their participation rate is lower than their country counterparts. Facilities and support services for multiple birth families are often lacking in rural areas; until recently, the Australian Multiple Birth Association (WA branch) (AMBAWA) only held regular meetings for parents of multiples in metropolitan regions. Many families who lived in country areas expressed their gratitude at being contacted and invited to participate, and were grateful for someone to talk to.

To get a definitive answer as to which method, or combination of methods results in the maximum response rate, more randomised studies examining all the various methods are needed. However, what is clear is that a combination of methods using questionnaire length, number and type of follow-up contacts, characteristics of likely participants and characteristics of staff should be taken into account when planning a questionnaire survey.

Endnote

- 1 Calculated using the total number registered on the WA Electoral roll, divided by the WA population (excluding visitors) aged 18 years or older at the last census (2001).

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References

- Anderson, L. S., Beverly, W. T., Corey, L. A., & Murrelle, L. (2002). The mid-Atlantic twin registry. *Twin Research*, 5, 449–455.
- Baker, L. A., Barton, M., & Raine, A. (2002). The southern California twin register at the University of Southern California. *Twin Research*, 5, 456–459.
- Bean, A. G., & Roszkowski, M. J. (1995). The long and short of it. When does questionnaire length affect response rate? *Marketing Research*, 7, 21–26.
- Bellamy, N., Duffy, D., Martin, N., & Mathews, J. (1992). Rheumatoid arthritis in twins: a study of aetiopathogenesis based on the Australian Twin Registry. *Annals of the Rheumatic Diseases*, 51, 588–593.
- Bergem, A. L. M. (2002). Norwegian twin registers and Norwegian twin studies — an overview. *Twin Research*, 5, 407–414.
- Boomsma, D. (1998). Twin registers in Europe: an overview. *Twin Research*, 1, 34–51.
- Boomsma, D., Busjahn, A., & Peltonen, L. (2002). Classical twin studies and beyond. *Nature Reviews Genetics*, 3, 872–882.
- Boomsma, D. I., Vink, J. M., van Beijsterveldt, T. C. E. M., de Geus, E. J. C., Beem, A. L., Mulder, E. J. C. M., et al. (2002). Netherlands twin register: a focus on longitudinal research. *Twin Research*, 5, 401–406.
- Boyd, N. F., Dite, G. S., Stone, J., Gunasekara, A., English, D. R., McCredie, M. R. E., et al. (2002). Heritability of mammographic density, a risk factor for breast cancer. *New England Journal of Medicine*, 347, 886–894.
- Casini, B., Pittaluga, M., & Parisi, P. (2002). Two Italian Twin registers for research in human biology and sport science. *Twin Research*, 5, 376–381.
- Cohen, D. J., Dibble, E., Grawe, J. M., & Polin, W. (1975). Reliably separating identical from fraternal twins. *Arch Gen Psychiatry*, 32, 1371–1375.
- Collins, R. L., Ellickson, P. L., Hays, R. D., & McCaffrey, D. F. (2000). Effects of incentive size and timing on response rates to a follow-up wave of a longitudinal mailed survey. *Evaluation Review*, 24, 347–363.
- Croft, M. L., Read, A. W., de Klerk, N. H., Hansen, J., & Kurinczuk, J. J. (2002). Population based ascertainment of twins and their siblings, born in Western Australia 1980 to 1992, through the construction and validation of a maternally linked database of siblings. *Twin Research*, 5, 317–323.
- Derom, C., Vlietinck, R., Thiery, E., Leroy, F., Fryns, J.-P., & Derom, R. (2002). The East Flanders prospective twin survey (EFPTS). *Twin Research*, 5, 337–341.
- Duffy, D. L., & Martin, N. G. (2001). Increasing the response rate to a mailed questionnaire by including more stamps on the return envelope: A cotwin control study. *Twin Research*, 4, 71–72.
- Duffy, D. L., Martin, N. G., Battistutta, D., Hopper, J. L., & Mathews, J. D. (1990). Genetics of asthma and hay fever in Australian twins. *American Review of Respiratory Disease*, 142, 1351–1358.
- Edwards, P., Roberts, I., Clarke, M., DiGuseppi, C., Pratap, S., Wentz, R., & Kwan, I. (2002). Increasing response rates to postal questionnaires: systematic review. *British Medical Journal*, 324, 1183–1191.
- Ellis, J. A., Stebbing, M., & Harrap, S. B. (1998). Genetic analysis of male pattern baldness and the 5 alpha-reductase genes. *Journal of Investigative Dermatology*, 110, 849–853.
- Glinianaia, S. V., Rankin, J., Wright, C., Sturgiss, S. N., & Renwick, M. (2002). A multiple pregnancy register in the north of England. *Twin Research*, 5, 436–439.
- Goldberg, J., Curran, B., Vitek, M. E., Henderson, W. G., & Boyko, E. J. (2002). The Vietnam era twin registry. *Twin Research*, 5, 476–481.
- Hansen, J., de Klerk, N. H., Croft, M. L., Alessandri, P. T., & Burton, P. R. (2000). The Western Australian Twin Child Health (WATCH) study: Work in progress. *Australian Epidemiologist*, 7, 16–20.
- Harrap, S. B., Stebbing, M., Hopper, J. L., Hoang, H. N., & Giles, G. G. (2000). Familial patterns of covariation for cardiovascular risk factors in adults. The Victorian Family Heart study. *American Journal of Epidemiology*, 152, 704–715.
- Hayakawa, K., Shimizu, T., Kato, K., Onoi, M., & Kobayashi, Y. (2002). A gerontological cohort study of aged twins: the Osaka university aged twin registry. *Twin Research*, 5, 387–388.
- Heath, A. C., Howells, W., Kirk, K. M., Madden, P. A. F., Bucholz, K. K., Nelson, E. C., et al. (2001). Predictors of non-response to a questionnaire survey of a volunteer twin panel: findings from the Australian 1989 Twin cohort. *Twin Research*, 4, 73–80.
- Hopper, J. L., Green, R. M., Nowson, C. A., Young, D., Sherwin, A. J., Kaymakci, B., et al. (1998). Genetic,

- common environment, and individual specific components of variance for bone mineral density in 10- to 26-year-old females: A twin study. *American Journal of Epidemiology*, 147, 17–29.
- Hopper, J. L. (2002). The Australian Twin Registry. *Twin Research*, 5, 329–336.
- Howell, S. C., Quine, S., & Talley, N. J. (2003). Ethics review and use of reminder letters in postal surveys: Are current practices compromising an evidence-based approach. *Medical Journal of Australia*, 178, 43.
- Jang, K. L., Livesley, W. J., & Vernon, P. A. (2002). The etiology of personality function: The University of British Columbia twin project. *Twin Research*, 5, 342–346.
- Kaprio, J., & Koskenvuo, M. (2002). Genetic and environmental factors in complex diseases: The older Finnish twin cohort. *Twin Research*, 5, 358–365.
- Kaprio, J., Pulkkinen, L., & Rose, R. J. (2002). Genetic and environmental factors in health-related behaviors: Studies on Finnish twins and twin families. *Twin Research*, 5, 366–371.
- Krueger, R. F., & Johnson, W. (2002). The Minnesota twin registry: Current status and future directions. *Twin Research*, 5, 488–492.
- Lamers, L. M. (1997). Medical consumption of respondents and non-respondents to a mailed health survey. *European Journal of Public Health*, 7, 267–271.
- Larroque, B., Kaminski, M., Bouvier-Colle, M.-H., & Hollebecque, V. (1999). Participation in a mail survey: role of repeated mailings and characteristics of nonrespondents among recent mothers. *Paediatric and Perinatal Epidemiology*, 13, 218–233.
- Levy, F., Hay, D. A., McStephen, M., Wood, C., & Waldman, I. (1997). Attention-deficit hyperactivity disorder: A category or a continuum? Genetic analysis of a large-scale twin study. *Journal of the American Academy of Child and Adolescent Psychiatry*, 36, 737–744.
- Mavis, B. E., & Brocato, J. J. (1998). Postal surveys versus electronic mail surveys. The tortoise and the hare revisited. *Evaluation and the Health Professions*, 21, 395–408.
- Morris, C. J., Cantrill, J. A., & Weiss, M. C. (2001). GP survey response rate: A miscellany of influencing factors. *Family Practice*, 18, 454–456.
- Mueller, B. A., McTiernan, A., & Daling, J. R. (1986). Level of response in epidemiologic studies using the card-back system to contact subjects. *American Journal of Public Health*, 76, 1331–1332.
- Page, W. F. (2002). The NAS-NRC twin registry of WWII military veteran twins. *Twin Research*, 5, 493–496.
- Parker, C. J., Dewey, M. E., on behalf of the TOTAL study group (2000). Assessing research outcomes by postal questionnaire with telephone follow-up. *International Journal of Epidemiology*, 29, 1065–1069.
- Sauerland, S., & Neugebauer, E. A. M. (2002). An experiment of mailing physician surveys on two different issues in joint or separate mail. *Journal of Clinical Epidemiology*, 55, 1046–1048.
- Skytthe, A., Kyvik, K., Holm, N. V., Vaupel, J. W., & Christensen, K. (2002). The Danish twin registry: 127 birth cohorts of twins. *Twin Research*, 5, 352–357.
- Spector, T. D., & MacGregor, A. J. (2002). The St. Thomas' UK adult twin registry. *Twin Research*, 5, 440–443.
- Spinath, F. M., Angleitner, A., Borkenau, P., Riemann, R., & Wolf, H. (2002). German observational study of adult twins (GOSAT): A multimodal investigation of personality, temperament and cognitive ability. *Twin Research*, 5, 372–375.
- Spry, V. M., Hovell, M. F., Sallis, J. G., Hofsteter, C. R., Elder, J. P., & Molgaard, C. A. (1989). Recruiting survey respondents to mailed surveys: Controlled trials of incentives and prompts. *American Journal of Epidemiology*, 130, 166–172.
- Stanley, F. J., Croft, M. L., Gibbins, J., & Read, A. W. (1994). A population database for maternal and child health research in Western Australia using record linkage. *Paediatric and Perinatal Epidemiology*, 8, 433–447.
- Stazi, M. A., Cotichini, R., Patriarca, V., Brescianini, S., Fagnani, C., D'Ippolito, C., et al. (2002). The Italian twin project: from the personal identification number to a national twin registry. *Twin Research*, 5, 382–386.
- Strassberg, M., Peters, K., Marazita, M., Ganger, J., Watt-Morse, M., Murrelle, L., et al. (2002). Pittsburgh registry of infant multiplets (PRIM). *Twin Research*, 5, 499–501.
- Sumathipala, A., Siribaddana, S., De Silva, N., Fernando, D., Abeyasingha, N., Dayaratne, R., et al. (2002). Sri Lankan twin registry. *Twin Research*, 5, 424–426.
- Sung, J., Cho, S.-H., Cho, S.-I., Duffy, D. L., Kim, J.-H., Kim, H., et al. (2002). The Korean twin registry — Methods, current stage, and interim results. *Twin Research*, 5, 394–400.
- Swanson, P. B., Pearsall-Jones, J., & Hay, D. A. (2002). How mothers cope with the death of a twin or higher multiple. *Twin Research*, 5, 156–164.
- Tambor, E. S., Chase, G. A., Faden, R. R., Geller, G., Hofman, K. J., & Holtzman, N. A. (1993). Improving response rates through incentive and follow-up: The effect on a survey of physicians' knowledge of genetics. *American Journal of Public Health*, 83, 1599–1603.
- Townsend, G. C., Aldred, M. J., & Bartold, P. M. (1998). Genetic aspects of dental disorders. *Australian Dental Journal*, 43, 269–286.
- Trouton, A., Spinath, F. M., & Plomin, R. (2002). Twins early development study (TEDS): A multivariate, longitudinal genetic investigation of language, cognition

- and behavior problems in childhood. *Twin Research*, 5, 444–448.
- Ward, J., Bruce, T., Holt, P., D'Este, K., & Sladden, M. (1998). Labour-saving strategies to maintain survey response rates: A randomised trial. *Australian and New Zealand Journal of Public Health*, 22, 394–396.
- Yang, H., Li, X., Cao, W., Lu, J., Wang, T., Zhan, S., et al. (2002). Chinese national twin registry as a resource for genetic epidemiologic studies of common and complex diseases in China. *Twin Research*, 5, 347–351.
- Zhu, G., Duffy, D. L., Eldridge, A., Grace, M., Mayne, C., O'Gorman, L., et al. (1999). A major quantitative-trait locus for mole density is linked to the familial melanoma gene *CDKN2A*: A maximum likelihood combined linkage and association analysis in twins and their sibs. *American Journal of Human Genetics*, 65, 483–492.
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