



Recruitment of Participants to a Familial Cancer Study: Balancing Privacy Protection and Research Quality

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Julia Slutsman, Ph.D.
Office of Human Subjects Research, NIH

Collaborators

- Louise Wideroff
- Barry Graubard
- Tim McNeill
- Anne Garceau
- Gordon Willis
- Marcia Dunn

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Disclaimer

The opinions expressed in this presentation are my own and do not necessarily represent the position or policy of the National Institutes of Health or the Department of Health and Human Services.

Familial Studies in Cancer Research

- A significant amount of cancer research requires the participation of family members as well as index-subjects
- Family based research is critical to the understanding of the genetic and environmental etiology of disease (Dorman et al., 1988)
- The success of family based research depends on the ability to identify, recruit, and enroll an unbiased sample of family members

Conduct of Familial Research

- Two common study designs for the conduct of familial research:
 - Model 1: Index subjects asked to provide personal health information about individual relatives
 - Model 2: Index subjects are asked to identify eligible family members who are then recruited (by researcher or family member) to participate in the research
- Fundamental trade-off between maximizing accrual and protecting privacy

Ethical Tradeoffs Between Recruitment Approaches

Pros:

- Control of relatives' health information is in participants hands

Cons:

- Index subjects and relatives can experience or perceive undue pressure associated with the recruitment process
- Difficulty assessing response rate
- Recruitment process may be more costly, less efficient

Subject Driven
Recruitment



(Beskow et al., AJMG, 2004, Parker et al., IRB, 1994)

Ethical Tradeoffs Between Recruitment Approaches

Investigator Driven
Recruitment



Pros:

- Higher response rates
- Accurate calculation of response rate
- Comparisons between respondents and non-respondents

Cons:

- Investigators collect information about family members without consent
- Potential for disclosure of private information about index subject, may cause distress

(Beskow et al., AJMG, 2004, Parker et al., IRB, 1994)

Summary of Existing Empirical Data

- All studies conducted on higher-risk populations
- Variation in preferences among recruitment methods
- Index subjects are willing to give researchers permission to contact relatives
- Little data about predictors of index subjects' willingness to give contact permission (to help with recruitment)
- Little data about characteristics of participating and non-participating relatives and their rates of enrollment in research

(Hull et al, *IRB*, 2004; Hadley, et al, *Archives of Internal Medicine*, 2003; Kreiger et al., *Annals of Epi*, 2001)

Family History Recruitment Study

- Secondary data analysis using data from the NCI Family Cancer History Validation Study conducted by NCI in 2000
- **Objective:** To assess the effects of an investigator driven recruitment strategy with respect to accrual in a population based cancer epidemiology study
- **Population:** Probability sample of the population of Connecticut (CT)
 - Subjects drawn from a random digit dial (RDD) sample of CT households
- **Eligibility:** (1) telephone in household, (2) CT residency, (3) age 25-64, (4) raised by at least one biological relative, (5) and at least one relative born or raised in the US or Puerto Rico

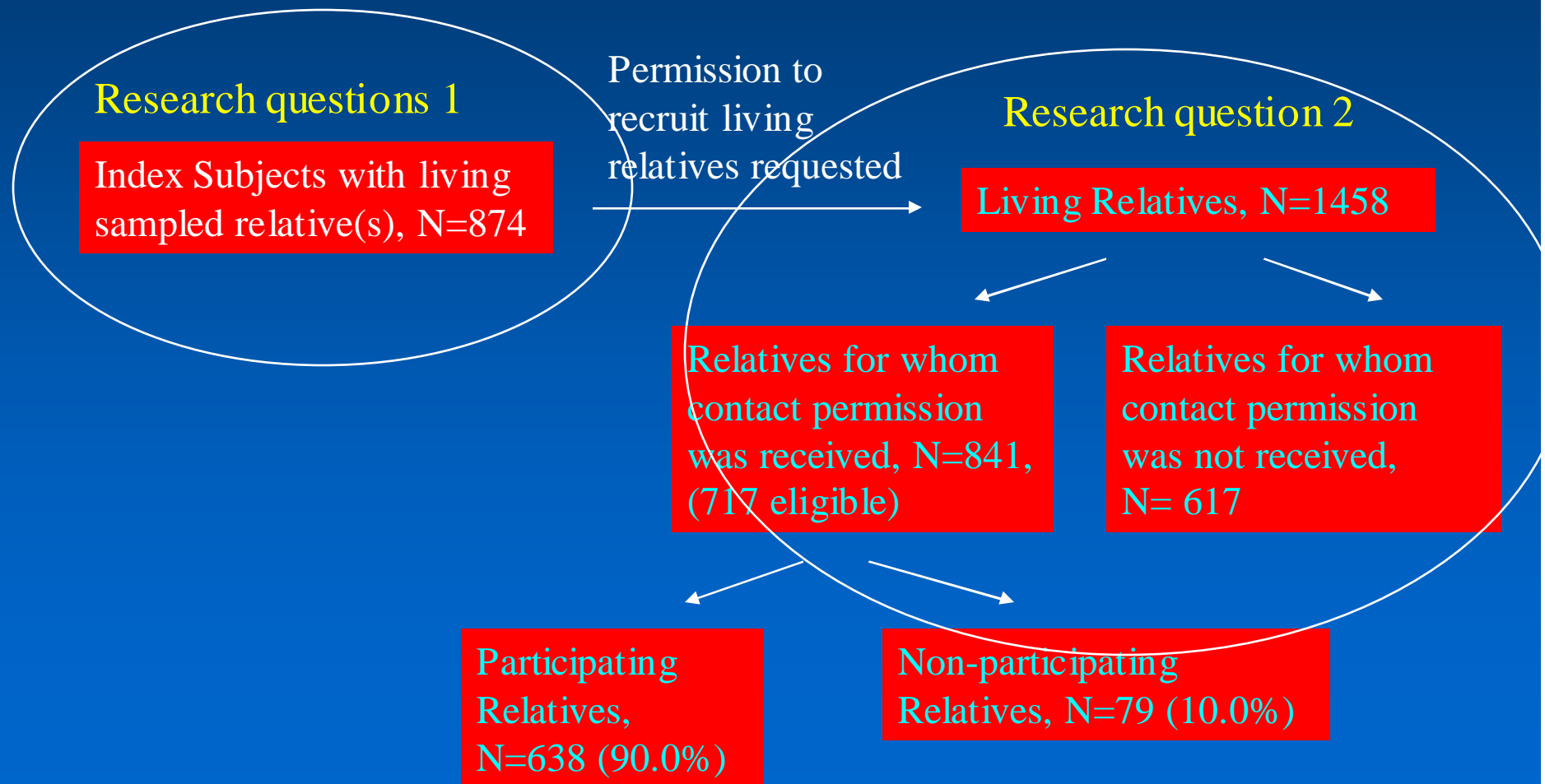
Family Cancer History Validation Study: Recruitment Strategy

- RDD used to identify eligible CT households from which a sample of index subjects (N=1380, RR=70.0%) was identified
- Index subjects asked to list all of their living and deceased relatives (N=28,629)
 - Investigators chose a sample of relatives for each subject
- Index subjects sent a workbook containing the names of selected relatives and a pre-paid phone card
- Index subjects who completed a second interview (N=1019) were asked to give contact permission for each relative
- Relatives for whom contact permission was received were sent an advance letter and contacted by research team

Family History Recruitment Study Samples

Population of Index Subjects

Population of Relatives



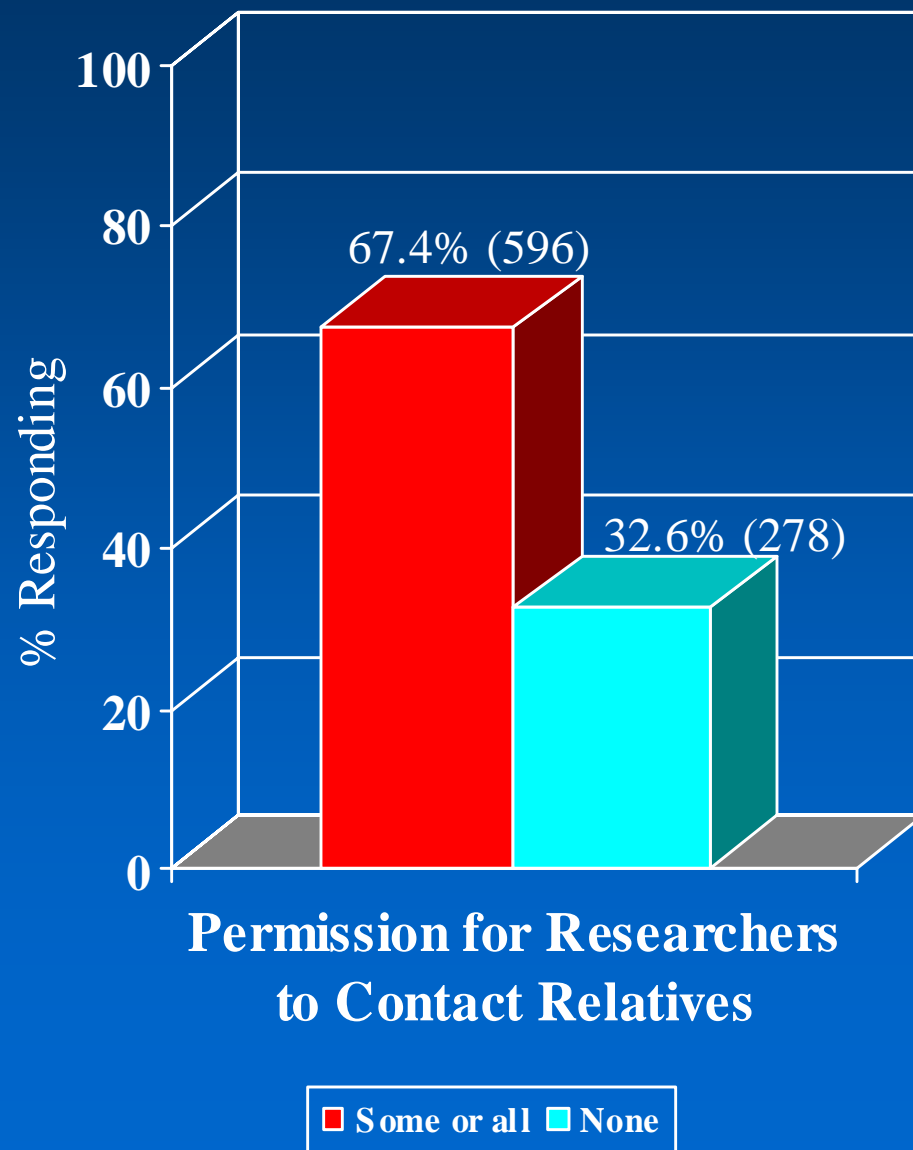
Research Questions

- Are index subjects' willing to give investigators permission to contact relatives?

Is index subjects' willingness to give investigators permission to contact individual family members associated with:

- relatives' demographic characteristics, and
- the closeness of the relationship between the index subject and relative?

Results: Willingness to permit contact



Research Question 2: Recruitment

Is index subjects' willingness to give investigators permission to contact individual family members associated with:

- relatives' demographic characteristics, and
- the closeness of the relationship between the index subject and relative?

Recruitment Study: Relatives Analyses

- Outcome Variable
 - Permission to contact individual relative (yes v. no)
- Independent Variables
 - Relative's Demographic Characteristics
 - Age, Sex, Cancer history
 - Index Subject's Demographic Characteristics
 - Sex, age, race, income, education , cancer history
 - “My (relative) and I are usually supportive of each other during difficult times” (yes v no)
 - “We can usually talk to each other about personal matters and problems” (yes v no)
 - Family cancer history
 - Contact with any relatives
 - Number of relatives for whom contact permission was requested
 - Family size
- All data from index subject self-report

Relatives' Demographic Characteristics (N=1458)

		Weighted % (sample N)	SE
Sex	Men	44.1% (550)	5.3
	Women	55.9% (908)	5.3
Age ¹	25-34	15.5% (11)	7.0
	35-44	14.6% (43)	3.7
	45-54	16.5% (205)	2.7
	55-64	16.2% (407)	1.7
	65 and over	34.8% (769)	3.4
Cancer history ¹	Positive	8.4% (230)	1.1
	Negative	89.9% (1204)	1.2
Biological Relationship ¹	First Degree Relative	50.4% (978)	4.9
	Second Degree Relative	48.1% (467)	4.7
Supportive Relationship ¹	Yes	75.2% (1156)	3.1
	No	23.7% (284)	3.1
Talk about personal matters ¹	Yes	68.8% (1055)	3.7
	No	29.7% (388)	3.6

¹ Excludes missing/DN.RF responses

Multivariate Logistic Regression Model: Contact Permission for Individual Relatives

		Total (n=874)	
		OR	95% CI
Intercept		0.11	0.03-0.37
Can talk to relative about personal matters**	Yes	5.13	2.66-9.92
	No	1.00	--
Relative's cancer history*	Positive	1.91	1.08-3.37
	Negative	1.00	--

¹This model is adjusted for: index subject's education level, # relatives for whom contact permission requested, contact with relatives, and family size.

*p <0.05, ** p<0.001

Conclusions

- This investigator driven recruitment strategy appears to be effective:
 - high proportions of index subjects willing to give researchers contact permission for their relatives (67.4%)
 - high rate of relatives participating in the study (90.0%)
- Investigator driven recruitment strategies in family studies may benefit from having this contact component built into the recruitment strategy
- Permission to contact individual relatives was associated with index subjects' ability to talk about personal matters with relatives as well as relatives' cancer status
 - source of bias in terms of which relatives researchers can contact
- Generalizability
 - Several limitations

