Special Article

Sampling Issues in a Regional Pediatric Practice-Based Network

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This paper explores various approaches to achieving representative and generalizable practice-based data. The data used were derived from original research conducted by the Pediatric Practice Research Group (PPRG), a research consortium including full-time staff at Children's Memorial Hospital and 132 practitioners in 35 Chicago-area practices in a variety of settings ranging from Chicago's inner city to rural northwest Indiana.

Sampling issues addressed include sampling frame, source (population or practice), geographical representation, seasonal representation, survey instrument design, socioeconomic diversity, and contact and response rates.

Key words. Data collection; selection bias; health surveys; sampling studies; research; health services research; practice-based research. (*J Fam Pract 1994; 38:415-421*)

Generalizability, "the extrapolation of findings from the specifics of the study setting to other settings,"[1] is one goal of clinical research. Hospital-based studies suffer from Berkson's bias,[2] ie, mild cases of a disease rarely reach the hospital. The result is that hospital-based research may not be generalizable to the broad range of patients and the severity of disease experienced by patients and seen by office practitioners.

Practice-based research has the potential to be generalizable to all practices, perhaps even to the general population.[3,4] A reference population is defined by person (age and sex), place (geographic location), and time (births within a particular time span) and includes all individuals eligible for a study population.[5]

The crucial first step in assessing the representativeness of each study sample is to determine whether it is representative of the reference population from which it was taken. The next step is to clarify to whom the study results can be generalized, ie, whether the results can be generalized beyond the reference population. To assess generalizability, a list of children and families recruited in each practice-based research study and characterized by age, sex, and socioeconomic status could be compared with general population statistics to determine whom the patients and families in the practice represent. However, this method is rarely feasible.

Short of this, various approaches can be used to maximize representativeness and generalizability by recruiting patients for practice-based research projects in as unbiased a fashion as possible. Methodological goals for sampling include geographical representation, seasonal representation, socioeconomic diversity, and high contact and response rates in each sample population (practice).

It is not always possible to design a study that meets all of these goals because strategies to achieve one goal may conflict with maximization of another. The aim of this paper is to use data from original practice-based research projects to illustrate various approaches to achieving representative and generalizable practice-based data.

The Pediatric Practice Research Group

The Pediatric Practice Research Group (PPRG) is an 11-year-old research consortium that includes full-time

Submitted January 31, 1994.

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ISSN 0094-3509

staff at Children's Memorial Hospital and 132 practitioners in 35 Chicago-area practices representing a variety of settings, from Chicago's inner city to rural northwest Indiana.[6,7] Studies by the PPRG are designed by PPRG and other hospital-based pediatricians in consultation with office-based pediatricians, managed by hospital-based PPRG staff, and reviewed at monthly meetings of hospital- and office-based PPRG members. Thirty-four of the 35 practices currently in the network have already participated in at least one study. Seven of the studies completed by PPRG are discussed below to illustrate lessons learned about sampling issues.

The Cholesterol Screening Study involved the prospective collection of family history and total cholesterol in 1005 prepubertal well children, with follow-up of those who had total cholesterol values >175 mg/dL (>4.55 mmol/L) (approximately 75th percentile) to determine whether family history is a sensitive screen for elevated low-density lipoprotein cholesterol (LDL-C).[8] Low-density lipoprotein cholesterol values ≥90th percentile for age and sex were identified in 88 children. Family history factors identified fewer than one half of the children found to have elevated LDL-C and did not selectively identify the most severely affected children.

The Infant Growth Study was a prospective cohort study (1 year enrollment, 1 year follow-up) of infant growth in 2574 healthy term infants at 10 PPRG practices.[9,10] Office staff obtained standardized anthropometric measurements, and parents completed a diet and health questionnaire at each health maintenance visit. Compared with current National Center for Health Statistics/Centers for Disease Control (NCHS/CDC) standards, PPRG infants were heavier through age 6 months and longer through 12 months. Revision of NCHS/CDC growth curves may be indicated.

The Hazard Exposure Study was a survey of 679 families with children over 3 years of age visiting participating pediatric offices during 1 month in each season of 1 calendar year to obtain information on age-specific exposure of Chicago-area children to amusement park rides, sleds, snow discs, bunkbeds, skateboards, fireworks, toboggans, and air guns.[11,12] This type of data may be used to refine anticipatory guidance and to correct injury rates for exposure.

The Firearm Survey addressed child exposure to firearms in households by means of a survey of 5233 parents. The parents attended 29 urban, suburban, and rural pediatric practices in Chicago, New Jersey, Houston, Utah, Georgia, Iowa, and South Carolina for health maintenance or sick-child care during a 1-week study period.[13] Firearm ownership and storage patterns were found to differ by practice location and family sociodemographic characteristics. Children were found to be at risk because of unsafe gun storage practices, offering an opportunity for physician intervention.

The Preschoolers Project was a prospective case-control study to assess how accurately pediatricians identify which preschoolers have emotional or behavior problems, or both,[14] and included 3876 children aged 2 to 5 years who were screened during a visit to one of 68 pediatricians. At the time of the visit, the pediatrician rendered an opinion about the presence of emotional or behavioral problems. Four hundred ninety-seven children who scored above the 90th percentile for behavioral problems on the Achenbach Child Behavior Checklist, along with children who had scored low and matched on age, sex, and race, participated in an intensive second-stage evaluation at Children's Memorial Hospital. The information from that evaluation was reviewed independently by two clinical child psychologists, who rendered an opinion about the presence of an emotional or behavioral disorder. Prevalence rates based on the psychologists' ratings were significantly higher than those of the pediatricians, suggesting that pediatricians need to enhance their recognition of preschooler behavior problems.

The Suburban Lead Study was a 3-month study of risk assessment and prevalence of elevated blood lead levels in suburban 1- and 2-year-old children.[15] Parents of children presenting for health supervision visits at pediatric practices completed questionnaires, and blood lead (BPb) levels were obtained for the children. Both questionnaire and an analyzable BPb level were available for 1393 subjects (79% of eligible subjects). Only 2.1% of the children were found to have a BPb level ≥10 μg/dL (≥0.48 μmol/L). The sensitivity of Centers for Disease Control and Prevention (CDC) risk assessment questions[16] was found to be 0.69. A revised risk assessment questionnaire, which includes the age of housing, may more accurately identify Chicago suburban children in need of BPb screening.

The Practice Characterization Study was a cross-sectional assessment of sociodemographic and practice mobility characteristics of 6811 families attending 28 PPRG practices over a 9-month period. The data collected are expected to clarify to what extent practice-based sampling approximates population sampling, define the population to which results of completed and ongoing PPRG studies are generalizable, and allow future studies to maximize generalizability by combining practices to match populations of interest.

Sampling Issues

Sampling Frame

Samples can be random or nonrandom, drawn from the general population or from office practices. Because a nonrandom sample involves the screening of fewer subjects, it requires less time to reach the target number. However, the sources of bias associated with nonrandom sampling techniques can be difficult to quantify and can result in imprecise information about bias, which complicates statistical corrections for bias in the analysis. In the Cholesterol Screening Study and the Suburban Lead Study, study participants included children of certain ages being seen for health maintenance visits. The eligible group was clearly identified, and participating offices were able to manage data collection.

Random sampling is unbiased but requires the screening of a large number of subjects. This can be accomplished by increasing staff in the practices, which requires additional funding, and screening a large number of practices. Random sampling within a practice can be accomplished by designating every *n*th eligible family for screening and/or recruitment, as in the National Ambulatory Medical Care Survey.[17] It also can be accomplished by using random number tables along with a numerical family identifier, such as a billing identification number, or by using marked envelopes to assign recruited subjects to either control or intervention status. An additional method of accomplishing random sampling is by randomizing recruitment sites or times. The Practice Characterization Study used a random number table to determine the rotation of screening assistants among practices and then recruited every family seen in the office during the designated recruitment shifts to avoid selection bias.

Source

The ideal reference population for pediatric practice-based research would be a random sample of patients in a random sample of all pediatric practices. Practices could be sampled either in a region (for a regional network such as the PPRG) or nationally (for a national network such as the Pediatric Research in Office Settings network of the American Academy of Pediatrics[18] or the Ambulatory Sentinel Practice Network[4]). However, not all practices and practitioners are interested in participating in research projects. The office size, configuration, or patient flow may not be conducive to the presence of a research assistant; physicians may not be interested in research, and office staff may not have the time or motivation to participate in data collection.

Various measures can be used to increase practice willingness to participate. The PPRG involves practitioners in discussing research ideas and elicits their participation in each step of the study. It also encourages practitioners to suggest modifications in study designs to account for crucial practice factors and in study protocol to make plans feasible in their offices. This improves study design and protocols because practitioners understand what ideas and study designs would work in their offices.

The Suburban Lead Study is a good example of this process. Although the State of Illinois recommends screening all children under 6 years of age, this volume of testing would have overwhelmed the practices. Therefore, screening was limited to children being seen for their 1- or 2-year-old checkup, the age range with peak incidence of BPb elevation.[19]

Geographical Representation

National samples have the advantage of providing the most generalizable data. However, unbiased sampling frames are difficult to create and implement. Procedures for quality-assurance monitoring of data collection and data entry in national studies have logistical and financial implications that affect implementation.

A regional sample has the advantages of ease of contact, decreased travel and training expenses for staff involved in the project, ease of data transfer to central computers for analysis, and potentially increased rapport between office staff and the centrally located project director.

Regional practice-based research networks have inevitable geographical limitations. The PPRG is geographically limited to the Chicago standard metropolitan statistical area and rural northwest Indiana. This constrains the generalizability of our results to pediatric practices outside the North Central standard metropolitan statistical areas and necessitates careful comparison of our samples with other settings wishing to "use" PPRG data, such as results from the Firearm Survey or the Suburban Lead Study. Generalizability is limited to similar practices unless oversampling is used to make the study sample representative of a wider population. Descriptive characteristics of each study's reference population can be compared with geographically comparable population data to assess degree of constraint.

Nonetheless, regional data are routinely used nationally, usually without recognition of the potential problems associated with this practice. An example is the widely used National Center for Health Statistics/CDC infant-growth charts based on a cohort from Yellow Springs, Ohio.[20]

Although the PPRG is a regional network, it does not always include all types of locales in studies (eg, the Suburban Lead Study). In other studies, a range of practice locations is necessary. The majority of the 10 practices who volunteered to participate in the Hazard Exposure Study were suburban, but there were enough urban and rural families to use area of residence as a variable in the analyses. Multivariate analyses compensated for the practice sampling frame. The Firearm Survey also relied on volunteer practices, but there were enough urban and rural families to derive significant results on the correlation between residence area and gun ownership.

Seasonal Representation

Seasonal variations in disease incidence or exposure patterns may lead to systematic errors in conclusions drawn from data collected over periods of less than 1 year. The ideal sampling technique to avoid seasonal bias would be continuous (complete or random) sampling over a 12-month cycle. However, the impact of this sampling on practice flow must be considered. Continuous sampling can impose a heavy burden on office staff, effectively block recruitment for other studies, and possibly result in reluctance to participate in future research. The Infant Growth Study involved six expected data collection points over the first 12 months of life. Even though obtaining the growth measurements was a routine procedure, the extra time required for the parent questionnaire and standardized measurements became a significant burden in some practices with an accumulating sample. The progressively increasing workload associated with questionnaire collection led some practices to stop enrollment early. However, variability in start dates for practice data collection prevented the overall enrollment from varying significantly by season.

Because of the impact of standardized procedures, such as growth measurements, on patient flow and staff time and effort, whenever feasible, we now use screening assistants in practices to handle the increased workload for high-volume or time-consuming studies conducted in the office setting, allowing longer data-collection periods without overburdening the practice.

Continuous sampling can result in repeat respondents. This problem skews results because repeat respondents tend to be younger, have chronic health problems, and be from larger families, and also because they may violate statistical assumptions of independence. Continuous sampling, therefore, requires survey techniques to efficiently identify duplicate forms. In addition to cover letters requesting that past respondents not complete a second form, we have used sociodemographic variable matching to identify duplication in study populations. In the Practice Characterization Study, a combination of practice, zip code, year of birth of oldest and youngest member of the household, and mother's education (highest year of schooling completed) was used to identify potential duplicate survey forms. Studies tracking subjects by name (Cholesterol Screening Study, Preschoolers Project, and Suburban Lead Study) have used this variable to identify repeats.

One alternative to continuous sampling is choosing 1 month to represent each of the four seasons and planning continuous sampling only during these study months. This sampling method, used in the Hazard Exposure Study, is less expensive and also considerably less intrusive than continuous 12-month sampling. Research staff were not available during the Hazard Exposure Study, however, and the combination of seasonal and age eligibility restrictions made it difficult for office staff to correctly distribute and collect survey instruments. Providing research staff during data-collection months could increase data completeness when using this study design.

In some cases, external constraints related to budget or personnel have resulted in data collection during only one season of the year (Suburban Lead Study, Cholesterol Screening Study). Although seasonal patterns that are not always consistent have been demonstrated for BPb levels,[21,22] they were considered less important than the availability of funding and support services, including specimen analyses by state laboratory, for a 3-month continuous data collection.

Survey Instrument

There is a conflict between the desire to gather large amounts of information and the limited time available for data collection in the office waiting room. Office practice is most conducive to short screening forms or surveys, as illustrated by the Hazard Exposure Study. The survey questionnaire, which included one page of questions for each of eight hazards, was completed primarily by middle-class respondents. With subsequent studies, reading level has been lowered to between fourth to sixth grade, and in-office data collection has been limited to two pages of questions with accompanying cover letter. This resulted in a better sociodemographic distribution of respondents in the Firearm Survey. Form completion has also been enhanced by an inviting format: the cover letter is printed on the first page of a "survey book" that opens to reveal two pages of questions on the inside.

Piloting survey instruments before beginning data collection increases the quality of data collected. A series of brief pilots of the Firearm Survey, the Suburban Lead

Table. Sociodemographic Characteristics of Pediatric Practice Research Group (PPRG) Practices

Variables	Pediatric Practice Research Group	Cholesterol Screening Study	Hazard Exposure Study	Infant Growth Study	Preschoolers Project	Firearm Survey†	Suburban Lead Study
Geographical location							
City	17*	3	3	4	9*	4	0
Suburbs	18*	4	6	4	9*	9	8
Semirural	2	1	1	2	2	1	1
Socioeconomic status							
Middle class	17	4	6	6	10	8	6
Indigent	5	1	0	1	1	2	0
Mixed	13	3	4	3	8	4	3
Race/Ethnicity‡							
>87% White	22	5	7	7	14	10	9
50% to 80% White	8	2	3	2	4	2	0
>90% Hispanic or black	5	1	0	1	1	2	0
Practices in each study, n	35	8	10	10	19	14	9
(% of PPRG)	(100)	(23)	(29)	(29)	(54)	(40)	(26)

**Some practices have urban and suburban offices.*
†Table does not reflect non-PPRG practice participants.
‡No practices with 81% to 86% white.

Study, and the Practice Characterization Study demonstrated problems with response categories and the wording of questions, which were resolved before formal data collection began.

Socioeconomically Diverse Populations

Even with a random sample of practices, diversity in the respondent population may be limited by sociodemographic factors, such as educational level and native language. Consideration must be given to survey instrument reading level and length and the availability of needed translations, which, in our case, was for Spanish-speaking people. In the Hazard Exposure Study, the complexity of the survey instrument limited the respondents primarily to educated members of the middle class. Modifications in length and content, with fewer open-ended response categories in the Firearm Survey and Practice Characterization Study, have facilitated participation of less well-educated respondents. We now place research assistants in offices where needed to assist illiterate respondents, and bilingual research assistants in offices attended by Spanish-speaking families.

Knowledge of a network's practice composition introduces the option of disproportionate stratified sampling to increase the sample from sociodemographic groups underrepresented in the network to more closely approximate the area population. The Preschoolers Project oversampled the participating inner-city minority clinic to increase representation of African-American families. The table illustrates some of the sociodemographic characteristics of practices participating in some PPRG studies. Refinement of practice descriptions and the inclusion of additional practice descriptors, such as family mobility and insurance status, will allow for more precision in practice sampling in future studies.

Contact Rate

The main factor limiting contact rate, which is the proportion of eligible families approached, is the availability of office staff to explain study participation to parents. Studies tracking routine procedures or surveys requiring minimal consent (the Infant Growth Study, Hazard Exposure Study, Firearm Survey, and Suburban Lead Study) have been completed entirely by office staff. We have sometimes successfully used retrospective evaluation of schedules or sign-in logs to assess contact rate, finding that office personnel were able to approach 94% of eligible children for the Suburban Lead Study. At other times, calculation of accurate contact rates was impossible without in-office staff.

For other studies, we have placed screening assistants in offices to explain the protocol, obtain parental consent, and administer survey instruments. For the Preschoolers Project, the study explanation, signing of consent forms, and completion of demographic information sheet and Achenbach Child Behavior Checklist took approximately 15 minutes per family.

Response Rate

It has been our experience that, in practice-based studies, enthusiasm of practitioners and staff is the most important factor influencing response rate, which is the proportion of invited subjects who choose to participate. If the *practitioners* are interested in the research topic, the staff transmit this enthusiasm to the collection of quality data. We work to maximize physician enthusiasm for studies by involving PPRG members in the choice of research topics that are of interest to them and the families they serve. The idea for the Suburban Lead Study was based on one practitioner's experience, which suggested that lead intoxication was an infrequent problem at his practice. With new BPb testing requirements in Illinois, many other pediatricians were also interested in BPb screening. *Parental* awareness of and interest in the research topic also influenced willingness to participate. Frequent news stories about lead exposure increased parents' awareness of the topic and influenced their willingness to consent to having blood drawn. Any requirement for invasive procedures or for return visits to the practice or to another site should be adequately explained, and the family should be offered appropriate financial compensation for their time, effort, and inconvenience.

In studies such as the Suburban Lead Study, office staff were responsible for all data collection. A 30-minute in-service training session on lead exposure and lead intoxication conducted at each office prior to the data collection period contributed to staff enthusiasm and increased their confidence about answering parental questions. Some orientation is provided at each office before each PPRG study, ranging in complexity from a brief drop-in visit (Practice Characterization Study) to a formal in-service training (Suburban Lead Study), and frequent contact with office staff is maintained throughout the data-collection period.

Enthusiastic staff are more likely to faithfully adhere to the standardized protocol, and their enthusiasm usually increases response rate. Adequate supplementation of the office staff with research assistants may be needed to increase contact rates and to facilitate consistent documentation of contact and response rates and characteristics of nonresponders.

The length of the survey form and the availability of dedicated research assistants can also influence response rate. A clear description of the study, including a cover letter on practice letterhead signed by practice doctors, has facilitated survey distribution, and personal attention at the time of enrollment seems to increase participation.

Incentives should be geared to the extent of participation and the specific needs of the population. Many in-office studies (such as the Infant Growth Study and Practice Characterization Study) can be performed with no incentive, whereas studies requiring extended participation (such as the Preschoolers Project) probably cannot. The request to travel to an alternate site for 3 hours of parent and child interviews for the Preschoolers Project was clearly a significant time commitment for the parent, warranting appropriate compensation. Because Chicago inner-city families find check cashing difficult, they prefer grocery store gift certificates when compensation is indicated.

Studies such as the Preschoolers Project, which required repeated contacts, can be affected by family mobility. At the time of recruitment, it is important to obtain alternative contacts for each enrolled family.

Discussion

Successful representative and generalizable practice-based research requires awareness of study design issues and possible approaches to building strong relationships with the participating practices. Practitioner involvement in every phase of study planning ensures that studies will be of interest to the practitioners, designed to make data collection feasible, and capable of generating staff enthusiasm. Careful design of survey instruments, translated forms, and bilingual staff allow for the participation of a sociodemographically diverse practice population. Sociodemographic characterization of practices allows for selection of the most appropriate reference population for each study.

Continued work by research networks will clarify remedies for several problematic design issues, including seasonality, nonrandom sampling, and assessment of generalizability.

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