

Multilevel Provider-Based Sampling for Recruitment of Pregnant Women and Mother-Newborn Dyads

Thomas J. McLaughlin, MA, MDiv, ScD,^a Onesky Aupont, MD, MPH, PhD,^a Claudia A. Kozinetz, PhD,^b David Hubble, MA,^c Tiffany A. Moore-Simas, MD, MPH, MEd,^a Deborah Davis, PhD,^d Christina Park, PhD,^e Ruth Brenner, MD, MPH,^e Deidre Sepavich, MBA,^a Marianne Felice, MD,^a Chantal Caviness, MD,^f Tim Downs, DEnv,^g Beatrice J. Selwyn, ScD,^h Michele R. Forman, PhDⁱ

abstract

OBJECTIVE: In 2010, the National Children's Study launched 3 alternative recruitment methods to test possible improvements in efficiency compared with traditional household-based recruitment and participant enrollment. In 2012, a fourth method, provider-based sampling (PBS), tested a probability-based sampling of prenatal provider locations supplemented by a second cohort of neonates born at a convenience sample of maternity hospitals.

METHODS: From a sampling frame of 472 prenatal care provider locations and 59 maternity hospitals, 49 provider and 7 hospital locations within or just outside 3 counties participated in study recruitment. During first prenatal care visits or immediately postdelivery at these locations, face-to-face contact was used to screen and recruit eligible women.

RESULTS: Of 1450 screened women, 1270 were eligible. Consent rates at prenatal provider locations (62%–74% by county) were similar to those at birth locations (64%–77% by county). During 6 field months, 3 study centers enrolled a total prenatal cohort of 530 women (the majority in the first trimester) and during 2 months enrolled a birth cohort of an additional 320 mother-newborn dyads. As personnel became experienced in the field, the time required to enroll a woman in the prenatal cohort declined from up to 200 hours to 50 to 100 hours per woman recruited.

CONCLUSIONS: We demonstrated that PBS was feasible and operationally efficient in recruiting a representative cohort of newborns from 3 diverse US counties. Our findings suggest that PBS is a practical approach to recruit large pregnancy and birth cohorts across the United States.



^aUniversity of Massachusetts, Worcester, Massachusetts; ^bEast Tennessee State University, Johnson City, Tennessee; ^cWestat, Rockville, Maryland; ^dUniversity of Louisville, Louisville, Kentucky; ^eNational Institute of Child Health and Human Development, Rockville, Maryland; ^fBaylor College of Medicine, Houston, Texas; ^gClark University, Worcester, Massachusetts; ^hUniversity of Texas School of Public Health, Houston, Texas; and ⁱUniversity of Texas, Austin, Texas

Dr McLaughlin conceptualized the manuscript and designed the analysis; acquired, analyzed, and interpreted the data; drafted the initial manuscript; and coordinated incorporation of the critical review and suggested revisions of all authors; Dr Aupont analyzed and interpreted the data, drafted the initial manuscript, and critically reviewed and edited the manuscript; Dr Kozinetz interpreted data, drafted the initial manuscript, and critically reviewed and edited the manuscript; Mr Hubble developed the study design, sampling procedures, and sample selection; acquired, analyzed, and interpreted the data; drafted the initial manuscript; and critically reviewed and edited the manuscript; Dr Moore-Simas interpreted the data and critically reviewed and edited the manuscript; Dr Davis interpreted data and critically reviewed and revised the draft manuscript; Dr Park analyzed and interpreted the data and critically reviewed and revised the draft and subsequent manuscripts; Dr Brenner developed the study design, interpreted the data, and critically reviewed the final manuscript; Ms Sepavich analyzed the data and critically reviewed and revised the draft and final manuscripts; Drs Felice and Downs drafted the initial manuscript and critically reviewed and revised the manuscript; Dr

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TABLE 1 Land and Population Characteristics of PBS Counties

	Harris, TX	Jefferson, KY	Worcester, MA
Land area in square miles	1703	1511	380
Population, 2011 estimate	4 180 894	801 227	746 906
Number of births, 2011 ^a	65 980	10 086	8772
Foreign born persons, %, 2007–2011	25.0	10.9	6.0
Non-English language spoken at home, %, age ≥5, 2007–2011	42.1	17.5	7.7
Median household income, 2007–2011	\$52 675	\$65 772	\$46 298
Persons below poverty level, %, 2007–2011	17.3	9.9	15.7

Source: <http://quickfacts.census.gov>.

^a Natality data.

As the National Children's Vanguard Study (NCS)¹ tested the enhanced household-based, provider-based, and direct outreach recruitment methods,^{2–4} a general consensus emerged among the NCS investigators that use of prenatal care providers rather than geographic-based secondary sampling units could be a more logistically efficient and cost-effective method for deriving a national probability sample of births in the United States.^{5–7}

In 2012, the provider-based sampling (PBS) pilot study was implemented by 3 study centers (SCs) to test an alternative sampling design and recruitment strategy using prenatal care providers. Unlike previous sampling and recruitment approaches, PBS had a multilevel probability sample frame of all prenatal provider locations from which a stratified random sample of provider locations was selected with probability proportional to size (the number of births attributable to provider location; PPS), and then within that provider sample subset, a sample of pregnant women was recruited.^{8,9} Like provider-based recruitment that had previously been studied for the NCS,³ PBS relied on access to study subjects through provider settings. However, unlike provider-based recruitment, PBS randomly sampled the entire set of providers, and within that provider sample, women were recruited using unique field methods that assured equal probabilities

of selection of study subjects from all women receiving care. As such PBS was designed as a multilevel, probability sample of pregnant women. Furthermore, to ensure representativeness of all births in the overall PBS sample, a birth cohort of women who had no prenatal care or who accessed care from a provider not listed on the sampling frame was recruited from maternity hospitals. The aim of the PBS was to develop a cohort representative of births in each county in a way that improved feasibility and acceptability and decreased costs compared with previous approaches. Although the National Institutes of Health ultimately decided that moving forward with the large-scale NCS was not feasible, this article describes a successful sampling and recruitment approach in 3 diverse counties. The objectives of the article are to describe the design, sampling frame creation, probabilistic sampling, and enrollment of the PBS pilot study and to discuss the diversity of approaches used to achieve these objectives by the three SCs.

METHODS

Study Counties

The PBS pilot study was conducted in Harris County, Texas; Jefferson County, Kentucky; and Worcester County, Massachusetts. Baseline descriptors of the 3 study counties differed by demographics and other characteristics of mothers as indicated in Table 1. This variation

provided real-world differences illustrative of geographically diverse areas of the United States and thus reveal patterns of PBS recruitment of women in a range of social and ecological conditions.

PBS Study Design and Multistage Probability Sampling

The overall PBS design developed 2 cohorts: a cohort of pregnant women sampled and recruited at prenatal provider locations over a 6-month period and a cohort of mothers and their newborns sampled and recruited over a 2-month period at the delivery hospital at least 12 hours postpartum. The PBS birth cohort included women who did not have prenatal care or who did not have care at the selected prenatal provider locations within the prenatal sampling frame. The PBS design had 2 fundamental approaches to the development of the cohorts in each county: (1) for the prenatal cohort, prenatal care provider locations were sampled and recruited followed by sampling and enrollment of pregnant women at their first prenatal care visit; (2) for the birth cohort, recruitment of hospitals was followed by enrollment of mother-offspring dyads postdelivery.

The PBS used a 2-level hierarchical probability sampling frame; at the provider, level the probability of selection was based on the number of births at each provider location from among all prenatal provider locations in the county and from locations outside the county if they delivered services to women residing in the study county. A sample of provider locations stratified by various characteristics (eg, percent Hispanic, percent Medicaid, and geography) was selected with PPS (based on the estimated number of births) as a representative set of provider locations. Within the selected sample of prenatal care provider locations, a probability sample of pregnant women was identified. In practice,

operationalizing the design for the prenatal care provider locations preceded a hospital-based sample for various reasons, including feasibility of initiating field operations and the administrative complexities of seeking permission and access to multiple organizational units within hospitals. Choice of the birth hospitals was based on a convenience sample because of the small number of hospitals in 2 of the 3 study counties. However, the sampling frame for identification and enrollment of mothers who recently delivered a newborn at the hospital used a probabilistic approach.

Development of the Sampling Frame and Selection of Prenatal Care Provider Office Locations

A list of all prenatal care provider locations in each county was developed to inform the number of births delivered by each provider location over a year and to create the sampling frame to select unique provider locations. A provider location, rather than each provider, was considered the unit of observation for the PBS and as such included private offices and public health clinics. Because a woman might seek prenatal care through providers other than obstetricians (eg, family medicine providers, midwives), the identification of providers went beyond 1 medical specialty. Moreover, provider locations were identified from multiple sources including birth certificates, Internet-based searches for providers, and lists of medical societies. To ensure completeness of the provider lists, providers were identified for women residing in the county and thus included offices inside and outside the county.

A Provider Frame Questionnaire (PFQ) was used in Worcester and Jefferson Counties to estimate the annual number of births (to serve as the measure of size) and other characteristics at prenatal care

TABLE 2 List and Frame Creation Process for Prenatal Care Providers

	Harris	Jefferson	Worcester
Compilation of prenatal care provider list			
Initial list of prenatal care providers	1000+	~60	145
Refined list of prenatal care providers	360	36	76
In-county providers serving county residents	286	33	45
Out-of-county providers serving county residents	74	3	30
PFQ administration			
Providers that completed PFQ and remained eligible (PFQ completion rate, %)	NA	31 (91)	58 (85)
Providers determined to be ineligible from PFQ responses	NA	2	8
Providers that refused/were nonresponsive to PFQ	NA	4	10
Total number of births in frame	67 095	13 468	10 579
2011 National Natality number of births	65 980	10 086	8 772
Ratio of Frame to Natality counts	1.03	1.33	1.21

NA, not applicable.

provider locations. The PFQ included items about provider location contact information, the numbers and types of providers practicing at the location, the number of patient visits, demographic composition of the patient population, and any affiliations of the provider office with a larger practice or group of locations. Thus, the PFQ had 22 questions that captured information either to inform the sample frame or to validate an already developed frame. In Harris County, birth certificates, not the PFQ, were used to compute annual birth counts per location and had other information to create the sampling frame. Table 2 lists prenatal care provider locations by county (>1000 in Harris County, ~60 in Jefferson County, and 145 in Worcester County) and, where relevant, indicates participation in PFQ efforts. Locations with <50 women seen for a first prenatal care visit per year were excluded from the sampling frame, reducing locations to 360, 36, and 76 provider locations for Harris, Jefferson, and Worcester counties, respectively. We stratified the frame by illustrative characteristics such as those described in the design section. Altogether, 16 to 17 provider locations per county were selected with PPS by sampling statisticians working alongside PBS teams with the original intent to enroll 250

women per SC over the course of a full year in the field.

Recruitment of Provider Office Locations

Staff at each NCS SC used multiple approaches to recruit selected prenatal provider locations including letters, in-person visits to the office, phone calls, and give-away items from the NCS. In some cases, previous research collaboration facilitated entry into the office. Typically each office had a gatekeeper who did or did not facilitate recruitment depending on the size of the practice, the personality of the gatekeeper, his or her role in the office, and other factors. Many locations required direct contact with the lead physician in the practice, clinic leadership, and/or medical and administrative leadership. In some locations, hospital corporations purchased practices during this study period, which required direct contact with corporate leadership and research personnel to gain entry. Iterative contacts were typically necessary to develop trust between the NCS staff and either the provider and office staff or the hospital staff. If a practice refused to participate or exhaustive contact attempts failed to result in a connection or approval for further efforts, the SC could request that the NCS Program Office replace the provider location.

After receiving provider approval to recruit at a location, the NCS SC team then worked on logistical issues to conduct the PBS at each specific location. Logistical issues included coordination and integration with office procedures, the identification of days and times when women attended first prenatal care visits, space for NCS data collectors, and prescreening methods described subsequently. During the process of establishing a base at the provider office, the NCS SC team would reach out to the office staff and engage ≥ 1 individuals who would become the “champion” of the NCS at the office. Once the PBS was established at a location, NCS SC staff continued efforts to maintain positive relations at the location as a result of diverse issues such as scheduling changes or holidays that altered the office/location routine.

Recruitment of Pregnant Women in Prenatal Provider Locations

Eligibility Criteria

A pregnant woman was eligible if she resided in the county, was 18 years or older, had a confirmed pregnancy, and was sampled at the first prenatal visit for this pregnancy. First prenatal visit was defined as the first time the woman received prenatal care at the selected location whether with physician, nurse, midwife, or other clinical care provider, provided that she had not received prenatal care for this pregnancy at any other sampled location. This definition served the dual purpose of establishing a unique event for each pregnant woman at which time she would be eligible for sample selection and to ensure enrollment as early in the pregnancy as possible.

Sampling of Women

On the basis of the estimated numbers of births at each location, a sampling rate was calculated for each provider location. Three sampling strategies were used: (1)

systematic (every Nth woman), (2) temporal (every Nth week in the data collection period), (3) 2-layered temporal (every Nth day in every Nth week in the data collection period). Strategies were discussed with each sampled provider location, and women were sampled according to the agreed on sampling strategy/rate. For example, at locations with the fewest births, every eligible woman was in the sample. At locations with a large estimated number of births, as few as 1 in every 24 women was in the sample. The sites were required, in general, to adhere to the assigned sampling rate for each location, but flexibility in the continued scheduling and other sampling activities was necessary to reduce the burden on the providers and to ensure continued provider engagement and access to the clinical site. Adjustments were made if the observed count of pregnant women sampled misaligned significantly from the expected counts based on the original estimate of annual births. It should be noted that on average, this estimate was most accurate in Harris County where birth certificate data were used as opposed to provider location responses to the PFQ questions on annual number of births per provider location. See Table 2, where the ratio of the frame to national birth counts was 1.03 for Harris County indicating fairly consistent MOS, whereas the ratios were 1.33 and 1.21 for Jefferson County and Worcester County, respectively indicating fairly large degrees of overcoverage.

Prescreening of Participants

Prescreening protocols for participant eligibility varied by provider location and county. Methods included provider location staff prescreening for pregnancy, first prenatal visit, county of residence, and/or age of women. All women who were prescreened as eligible were either approached by data collectors or by provider staff in 1

county to complete the eligibility screener. The screener had questions about race, education, ethnicity, and language of the women to match staff assignments for consent and follow-up to linguistic and other needs and to compare women who did or did not enroll.

Selection and Recruitment of Birth Hospitals

Within each county, all hospitals and birthing centers were identified using the expertise of the NCS team, birth certificates, publically available resources, personal communications, and networking. In each county, 3 maternity hospitals were purposefully selected. In Harris and Worcester counties, the selection reflected variability in hospital location (urban/suburban/rural), annual birth count, and hospital type (public/private). In Jefferson County, 2 of the 4 hospitals were associated with 1 health care system; thus, 1 of these 2 hospitals was selected along with the remaining 2. Negotiations with the selected hospitals were held with hospital administrators (eg, Chief Executive or Medical Officer, Director of Nursing). Directors of Labor and Delivery, the Newborn Nursery, postpartum units, and institutional review board (IRBs). Two of 3 selected hospitals participated at 2 of the counties, and 3 hospitals participated in the third county. Of note, the nonparticipating hospital did not decline participation; rather, time ran out to complete hospital recruitment for this pilot study. Some hospitals and provider locations deferred to the *Eunice Kennedy Shriver* National Institute of Child Health and Human Development IRB for regulatory approval, whereas others required their own IRB review and approval. Health Insurance Portability and Accountability Act waivers were required to access hospital medical records.

Sampling, Identification and Recruitment of Mother-Offspring Dyads at Birthing Hospitals

Sampling

Operational efficiency guided the sampling of new mothers; each hospital rotated enrollment in sample once every 3 weeks. Eligibility criteria for the new mother included the following: (1) age ≥ 18 years, (2) county of residence, and (3) having no prenatal care at all or at a provider location not listed on the sampling frame for the prenatal cohort.

Identification of sampled women.

NCS SC staff used hospitals' electronic medical record systems and/or notification by hospital staff of all women who delivered a live birth during the sampled week. Subjects were prescreened for eligibility before being approached by NCS SC staff for screening, consent, and enrollment. To ensure that eligible births were not missed during the sampling period, a staff member confirmed births during the sampling period by reviewing delivery logs.

Recruitment

Beginning 12 hours postpartum, study staff alone or accompanied by a hospital neonatal nurse approached prescreened women at their bedside on the hospital maternity ward. An overview of the NCS was provided and an eligibility screener conducted with women interested in participation. Eligible women were asked to complete the consent and a birth interview. When circumstances prevented administration of the screener, consent, and/or interview, a follow-up visit was scheduled during the hospital stay; otherwise the woman was either a "loss" to recruitment or the birth interview was completed at home within 10 days postdelivery.

Overall Recruitment Target

The target for the PBS recruitment over a year of the pilot study was to

enroll 250 births per county. From each county, it was anticipated that ~200 eligible pregnant women and 160 eligible new mothers would be identified and enrolled in the study. Assuming a 70% average enrollment rate, 250 newborns were anticipated to be enrolled from a total of 360 eligible women in each county per year.

Data Sources and Quality

Data collected from provider offices and from potential and enrolled participants were transmitted to an NCS central data repository. Data that were collected on paper were entered into the electronic database using a double entry system at a SC. Complete entry and transmission of all field data into the central database, and the subsequent implementation of quality control checks resulted in the analytic files for this manuscript. Tracking of major study events including sampling, screening, consenting, and data collection was carefully managed through weekly calls between SCs and the coordinating teams until recruitment efforts ended July 30, 2013. National Natality data were obtained from the National Center for Health Statistics, Centers for Disease Control and Prevention.

Time Management Measures

SCs reported hours spent on management and data collection tasks on a weekly basis using a standardized format. Data were aggregated into 2 broad categories of "management activities" and "data collection." Management tasks included overall PBS study management by team leadership, study coordination, hospital outreach, provider outreach, IRB management, site office management (data and informatics management, subcontract oversight, geographic information systems specialist activities, PBS frame building, provider and hospital recruitment,

and PFQ administration). Data collection tasks included PBS participant eligibility screener and recruitment, informed consent, first and second pregnancy and birth visits, phone center infrastructure and interviewing, and study mailings. Each management or data collection task was operationalized using standardized definitions.

RESULTS

Prenatal Care Provider Location Recruitment

To achieve the target enrollment of pregnant women at provider locations in each county in a year, 16 to 17 prenatal care provider locations were recruited from each of the 3 county SCs. Table 3 presents the results of the provider location recruitment including refusals, the use of substitute locations and ineligible locations. The range in the total numbers of selected providers differed by site with 32 in Harris, 25 in Jefferson, and 22 in Worcester Counties. All sites experienced refusals or ineligible provider locations, the reasons for which varied but included the following: the provider or group in the location no longer provided obstetric services; the provider location had moved; or the office chose not to participate. In these instances a replacement provider location was selected. The final participation rates of eligible provider locations were: 64% in Harris, 71% in Jefferson, and 76% in Worcester counties. An important distinction and possibly contributing factor in these results is that with the use of the PFQ in the frame-building process, Worcester and Jefferson Counties had established contact with all sampled provider locations, whereas Harris County had not because of the use of birth certificates to build their provider location frame.

Prenatal Cohort Recruitment

Enrollment of pregnant women at the provider location occurred within 5 to 7 months across the 3 counties. Figure 1 displays rates of enrollment of pregnant women by county. When the cumulative average number of prenatal cohort members reached the target number, the prenatal cohort recruitment was terminated in July 2014. Table 4 presents the recruitment process starting with all patient visits in the provider office sample frame from which pregnant women were sampled, screened, and consented. The percentage of completed screeners varied by site with the highest rate of completion in Worcester County, then Harris, and finally Jefferson County. The low screen completion rate in Jefferson County was largely attributed to a logistical barrier that most providers in that county did not allow NCS SC staff to approach potential participants. Under a strict interpretation of the Health Insurance Portability and Accountability Act guidelines, potential participants had to voluntarily approach NCS SC data collectors. The consent rates ranged from 62% in both Harris and Worcester Counties to 74% in Jefferson County, whereas the numbers of completed pregnancy and birth visits varied by county and by gestation age at consent. The distributions of gestational age at recruitment differed by site with the highest percentage enrolled in the first trimester occurring in Worcester County (76%) followed by Jefferson (54%) and then Harris (39%) Counties (Fig 2).

Recruitment of Mother-Newborn Dyads at Hospitals

After recruitment of the prenatal cohort, the SCs recruited mother-newborn dyads to enroll women without prenatal care or outside the selected provider sampling frame. The PBS protocol imposed eligibility

TABLE 3 Number of PLs Selected and Recruited

	Harris	Jefferson	Worcester
Original PLs selected, <i>n</i>	16	17	16
Agreed participation	9	12	12
Refused participation	5	4	3
Determined ineligible	2	1	1
Participation rate among original PLs selected that were eligible, %	64	75	80
First substitute PLs selected, <i>n</i>	7	5	4
Agreed participation	2	2	2
Refused participation	2	3	2
Determined ineligible	3	0	0
Second substitute PLs selected, <i>n</i>	9	3	2
Agreed participation	5	3	2
Refused participation	2	0	0
Determined ineligible	2	0	0
Total PLs selected, <i>n</i>	32	25	22
Total agreed participation	16	17	16
Total refused participation	9	7	5
Total ineligible	7	1	1
Final participation rate among eligible PLs	64%	71%	76%

PLs, provider locations.

TABLE 4 Prenatal Care Cohort Recruitment

	Harris	Jefferson	Worcester	Total
Provider locations recruiting	16	17	16	49
No. of patient visits across all provider locations in sample frame	23 560	27 122	23 024	74 706
Women sampled and prescreened for eligibility screener	858	971	528	2357
Screeners completed (completion rate, %)	372 (43)	213 (22)	359 (68)	944 (40)
Eligible for consent based on eligibility screener (consent eligibility rate, %)	326 (88)	189 (89)	302 (84)	817 (87)
Consents completed (consent rate, %)	203 (62)	139 (74)	187 (62)	529 (65)
Pregnancy visit 1 completed	167	132	174	473
Pregnancy visit 2 completed	94	108	146	348
Birth visit completed	74	108	80	262

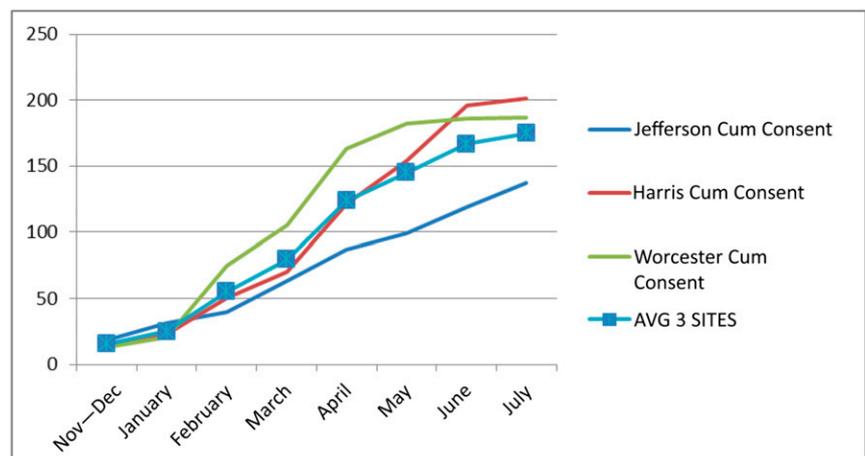


FIGURE 1

Cumulative Counts of Women Enrolled in Prenatal Cohort, by County and the Average Number for the 3 Counties

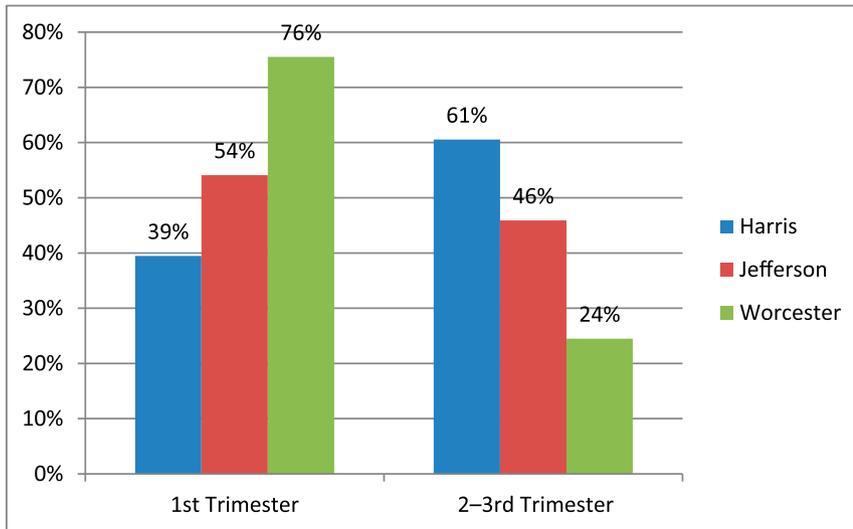


FIGURE 2 Percent of women by gestational age in trimester at time of consent in prenatal cohort. Cum, cumulative.

TABLE 5 Birth Cohort Recruitment

	Harris	Jefferson	Worcester	Total
Hospitals participating	2	3	2	7
No. of births in the hospital sample frame	692	356	842	1890
Women sampled and prescreened for eligibility screener	366	234	299	899
Screeners completed (completion rate, %)	201 (55)	61 (26)	247 (83)	509 (57)
Eligible for consent based on eligibility screener (consent eligibility rate, %)	182 (91)	41 (67)	229 (93)	452 (89)
Consents completed (consent rate, %)	117 (64)	27 (66)	177 (77)	321 (71)
Birth visit completed	111	26	173	310

challenges for recruitment including contacting women ≥ 12 hours postpartum and prohibiting initial contact after hospital discharge. Despite these restrictions, between 64% and 77% of eligible women were consented by NCS SC staff as described in Table 5.

Comparison of Consented Participant Characteristics Relative to the County

Table 6 presents sociodemographic profiles of the PBS prenatal and birth cohorts who consented to be in the study and contrasts it with 2011 county-specific birth data and the American Community Survey data. The most striking differences indicate that the PBS cohort overrecruited mothers who were Hispanic, aged

≥ 35 , and unmarried; had a high school education; and reported family incomes in the lowest quartile.

Proxy Measures of Resource Utilization

PBS activities for a variety of tasks were tracked and classified and depicted in Fig 3 as total hours required per enrolled participant. There was variation in hours across the 3 SCs; however, in all 3 SCs, the number of hours per enrollee declined over time, presumably indicating decreasing hours devoted to start-up activities and efficiencies over time.

DISCUSSION

The PBS pilot study had a multilevel probability sample of provider

locations to identify and recruit 530 pregnant women at the first prenatal visit, and 320 mother-newborn dyads from delivery hospitals, across 3 diverse counties in the United States. The study demonstrated that this sampling design and recruitment approach was feasible over a window of 6 rather than the originally targeted 12 months for the prenatal cohort and over a brief window of 2 months for the birth cohort. The PBS design began with an exhaustive list of all prenatal care providers from which a sample was drawn stratified by provider location characteristics. The list of prenatal care provider locations was developed and cross-verified from diverse sources such as birth certificates, the Internet, and a provider frame questionnaire. Over the course of the PBS study, the time required to enroll a woman over the recruitment declined from an initial start-up requiring time intensive activities to what appears to be reduced use of resources as indicated in hours per enrolled woman.

Over half of the prenatal cohort was recruited in the first trimester of pregnancy. Yet enrollment by trimester varied by county perhaps because of county-specific differences in the proportion who seek early prenatal care. For the earlier NCS alternative recruitment approaches, all of which used a geographic sampling approach for the second stage of sampling, enrollment of pregnant subjects was, on average, $\sim 20\%$, 40% , and 40% for the first, second, and third trimesters.¹⁰ Early recruitment underscores the value of PBS for study of early maternal and fetal exposures on future child health.

The rationale for provider-based sampling as an approach for recruitment of a large, prospective pregnancy cohort design was informed by previous survey and statistical sampling research that was summarized previously.^{8,9} This design applied to the NCS was presented by Belanger et al,

TABLE 6 Percent Distribution of Women Enrolled in PBS Prenatal and Birth Cohorts Versus County Level Data by Demographic Characteristics

Demographic Characteristics	Harris County			Jefferson County			Worcester County		
	Enrolled		2011 Natality	Enrolled		2011 Natality	Enrolled		2011 Natality
	n	%	%	n	%	%	n	%	%
Ethnicity/race									
Hispanic	217	68	51	51	31	7	77	21	14
Non-Hispanic white	18	6	25	52	31	63	215	59	72
Non-Hispanic black	68	21	18	41	25	26	21	6	6
Non-Hispanic other	16	5	6	22	13	4	52	14	7
Age Range, y									
<25	93	29	34	38	23	34	63	17	24
25–29	92	29	27	48	29	29	97	27	28
30–34	79	25	24	48	29	25	120	33	30
35+	56	18	15	32	19	12	83	23	18
Marital status									
Married	102	32	55	62	37	52	218	60	63
Not married	218	68	45	104	63	48	147	40	37
Education									
Less than high school	106	33	31	43	26	17	37	10	9
High school	108	34	22	50	30	20	93	25	27
Some college	73	23	24	38	23	32	95	26	25
College degree or higher	31	10	23	35	21	31	140	38	39
Income ^a									
<\$30,000	193	65	31	97	61	40	121	34	23
\$30,000–\$49,999	65	22	17	31	20	19	55	16	10
\$50,000–\$99,999	23	8	29	19	12	18	95	27	17
≥\$100,000	14	5	24	11	7	23	82	23	50

Women with unknown demographic characteristics are excluded from computing percent distribution.

^a County population reference data for demographic characteristics are from 2011 natality data except for income, which is available from 2012 American Community Survey data.

who opined that household-based sampling may be prohibitively expensive and unable to recruit sufficient numbers of pregnant women.⁵ An attractive alternative design consisted of multistage probability sampling through care locations in which all persons in the county have the same chance of random selection through medical providers and in which subjects are selected with known probability. A similar approach previously used by the Agency for Health Care Research and Quality to identify a national probability sample of HIV patients has been described, principles important in the success of that study and that were affirmed in the PBS research.^{8,9}

The principles driving implementation of the PBS were flexibility, choreography, and trust. Because the 3 PBS counties varied by geography, cultural, and social milieu, number of providers, population density and demographics, previous engagement with research, and existence of social networks, engagement and field activities were implemented dynamically and flexibly. These approaches took into account language and cultural diversity, the hierarchical or nonhierarchical structure and decision-making of the practice, the choreography of integrating NCS activities into the practice with minimal perturbation for the patient or provider, and clear assurances that the location could withdraw from study at any point should the research be viewed as impeding clinical flow.

Although only 3 in number, the SCs had quite diverse historical relationships between the provider community and the SC. As such, an SC and a provider community could have a long-standing social network that facilitated communication and sharing of values. In contrast, the SC and provider might have had a previous research collaboration

or even an adversarial experience that would require the appropriate “influentials” to help garner support for the study, beginning with buy-in from the key decision-makers and recruitment of the providers.¹¹ Although many participating providers had agreed to the study for altruistic reasons (eg, benefits to future generations of children), there was deep appreciation by study staff that clinical care was paramount.

The PBS succeeded in recruiting prenatal and birth cohorts after delivery under the direction of the National Institutes of Health through contractual arrangements and in the environment mandated by a congressional act. Yet unlike the multilevel sampling for prenatal care providers, selection of the small number of hospitals for birth cohort recruitment was purposeful. The engagement of hospitals was complex, involving multiple administrative units, and sensitivity to recruitment at hospitals with recent changes in leadership or accreditation. For example, academic medical centers were accustomed to having research conducted on the labor and delivery floors, and hence, nursing and clerical staff did not need extensive orientation. On the other hand, subject recruitment and data collection in such venues often competed with other ongoing research by academic faculty. In community hospitals, there was limited competition with existing research, but the staff required more education about following study protocol and the need for consistency and quality control. For-profit hospitals were concerned about the effect of the PBS on staff productivity and fiscal consequences. Recognition of the competing risks for birth cohort recruitment at diverse hospitals occurred through venues including debriefing sessions on the study,

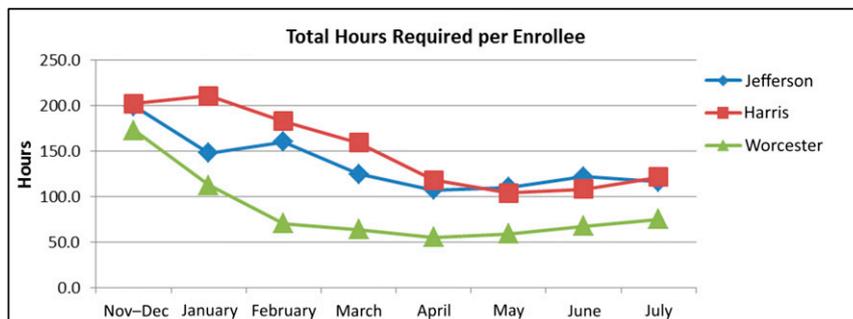


FIGURE 3

Total hours of administrative/management and field activities/data collection required per consented woman in the prenatal cohort.

one-on-one meetings with hospital leadership to ameliorate concerns about recruitment of new mothers postdelivery and submission of informational materials to organizations aligned with hospitals.

Several challenges were unanticipated. A major barrier to implementation of the PBS was the need to develop a list of all provider locations to formulate a frame for representative probability sample. This list had practitioners from diverse disciplines beyond obstetrics and nontraditional practitioners found in immigrant communities. In addition, listed practitioners often had multiple addresses but delivered different services at the various locations, such as birth control or fertility services at 1 site and prenatal care at another. These ambiguities were addressed site-specifically, ranging from a review of birth certificate data to mailing of provider questionnaires, all of which served to enumerate the annual number of live births at each provider location. Additionally, during birth cohort recruitment, time for consent and administration of the birth interview occurred after delivery and competed with visits by family and friends, by lactation consultations, and other activities for new mothers. Among specific ethnic groups, fathers played a major role in consent, and therefore consent would take place with the father present.

CONCLUSIONS

The PBS pilot study documented the ability to enroll a probability sample of prenatal care locations. It demonstrated that operations of prescreening, eligibility screening, and recruitment of pregnant women at first prenatal visits in provider locations is feasible, as is the screening and recruitment of new mothers at birth hospitals, to reach those who are not eligible in the prenatal frame. Finally, resource utilization, as quantitated by total administrative and field management activities, ranged from ~50 to 200 hours per enrollee depending on the site and the stage of the study and declined over time. These findings indicate that the PBS strategy may be a useful and efficient strategy to recruit pregnancy and birth cohorts representative of the source population.

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Community Advisory Boards gave freely of their limited time and suggested ways in which we could access study locations. We thank them for their participation. We also acknowledge the many staff members at all 3 sites who worked with deep dedication on the project. Finally, we are saddened to have lost Kathleen Belanger, our NCS colleague

from the Yale University School of Public Health, who died in the winter of 2013 and who had taken the lead in proposing PBS as a recruitment strategy for large, complex cohort studies. We dedicate this article to her memory and express our deep gratitude for the experiences of her intelligence, research abilities, and dedication.

ABBREVIATIONS

IRB: institutional review board
NCS: National Children's Study
PBS: provider-based sampling
PFQ: Provider Frame
Questionnaire
PPS: probability proportional to size
SC: study center

Caviness critically reviewed and revised the draft and final manuscripts; Dr Selwyn drafted the initial manuscript and interpreted the data; Dr Forman drafted the initial manuscript, interpreted the data, and critically reviewed and revised subsequent manuscripts; and all authors approved the final manuscript as submitted.

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Address correspondence to Thomas J. McLaughlin, ScD, Department of Pediatrics, University of Massachusetts Medical School, 222 Maple Ave, Shrewsbury, MA 01545. E-mail: thomas.mclaughlin@umassmed.edu

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