Expressing Your Priorities for the NCS

R.T. Michael

This exercise is intended to provide the FAC some sense of the prevailing priorities about the NCS that might guide judgments regarding the sampling design of the study. The exercise should not require more than ten minutes of your time; it will be more successful if you respond with your initial instincts rather than ponder the implicit complexities of the study before you respond and if you do not attempt "to game" the outcome by overstating your real views to influence the averages. The exercise has two separate parts; both explore the same few issues and the repetition is intended to give different perspectives on essentially the same few issues that may affect the sampling design of the NCS.

PART 1: In this exercise, assume that reasonably sensible decisions will be made about all the issues listed, since all are undoubtedly important to the success of NCS. The question for you is where you place your greatest interest in behalf of the study. To indicate your priorities, you have 100 points to allocate to any one or any combination of the seven domains listed below. Put your points where your passions lie.

There are seven domains here, described as follows:

I am most interested in or passionate about:

E the study's insights about one or a few of the **environments** that are a focus of NCS

O the study's insights about one or a few of the child **health outcomes** of focus of NCS

M the study's **mechanisms** (medical, familial, social…) that connect the environments and outcomes of focus in the NCS

L the study's **long-term research potential**, such as focus on selecting issues in infancy that are most likely to have payoff in adult health.

I the study's insights for the **immediate future**, those pertaining to the pregnancy and the neonatal period.

G the **generalizability** of the study's results to a wide spectrum of children

S the insights or results that pertain to **specific or particular groups of children**, such as those in poor families, African-American, or those served by medical centers of excellence.

E_____
O_____
M_____
L_____
I_____
G_____
S_____
Total: 100.

[As an example, if you think a pivotally important focus that will be a big factor in the ultimate payoff from NCS should be the findings about the effects on pregnancy of certain chemical environmental insults on all children, you might allocate 30 points to E, 20 points to O, 40 points to I and 10 points to G.]

PART 2: Here you are confronted with four separate pairs and for each of the four, please indicate where you stand, in terms of the trade-offs to be made by NCS. These four choices are independent of each other. Express your priority on each separate issue by placing an “X” along the line of each of the four continua.

2a: Hypothesis-driven v serendipity in NCS potential

Here, the issue is not how to craft a particular investigation with the data, it is instead how to think about the nature of the data to be collected. If you think the NCS's potential lies mostly with the specified "core" hypotheses, put your priority for the hypothesis end of the continuum which will imply a heavy weight to capturing the specific pieces of information critical to those core hypotheses. If, on the other hand, you think the NCS's potential lies mostly with the omnibus character of the wide-ranging data set that will provide opportunity for inquiries not currently envisioned, then express your priority for the "serendipity" end of this continuum which will imply placing a heavy weight on capturing information more broadly so those research opportunities that come from unanticipated changes in environments and new knowledge can be exploited.

Hypothesis Driven Inquiry — Serendipity Enhanced Inquiry

1 2 3 4 5 6 7 8 9 10

2b: Generalizability

Here, the issue is how important it is to you that the findings from the NCS are applicable to *at least fifty percent* of *all* children born in the U.S. in the time interval of the NCS's selection of live births for the NCS. (Some sampling schemes yield samples that can project to large populations, other schemes yield samples that project to none or to few others than those actually in the group of observations. The question here is how widely do you think it is important for the NCS findings to be applicable.)

Not important to me — Somewhat Important — Of Paramount Importance to me

1 2 3 4 5 6 7 8 9 10

2c: Universality of the key findings.

Some “findings” from the NCS are likely to apply to all children because those findings are universal, as are chemical reactions and many in-the-body environment-outcome mechanisms. Other likely “findings” from the NCS are probably dependent on the circumstances and behavioral responses that accompany the exposure to those environments, so these “findings” are not universal but instead highly context specific. The sample of pregnancies or children needs to be consistent with the judgment about how universal the important findings from NCS are: if those key findings are in-the-body or chemical relationships, for example, it may not matter who the observations are or whether they “represent” a larger population of children, but if those key findings involve social circumstances or varied responses, then that lack of universality calls for a probability sample. So this continuum asks you how invariant, universal you think the NCS’s key findings probably are.

Most Key NCS findings are Universal

Most Key NCS Findings are **NOT** universal

1 2 3 4 5 6 7 8 9 10

2d: The Trade-off of data precision and generalizability of NCS Findings

Here, like exercise 2b, you are asked to think about the population of children to whom you think the NCS findings should apply, but here the “trade-off” of generalizable and data quality is confronted. It would of course be ideal if the findings pertained to “all children” and if the data in the data set were perfectly measured, captured, and characterized, but both these ideals will be sacrificed by any real study done at any realistic expense. Thus the trade-off this exercise asks you to confront. The topics you hold most dear will influence your choice here.

quality, detail, precision of *measurement* captured data is of highest priority to me

generalizability to a wide known, population of children is of highest of priority to me

1 2 3 4 5 6 7 8 9 10

Thank you.

Response to Final Report from the National Children's Study Sampling Design Workshop

Fertility & Early Pregnancy Working Group
June 9, 2004

The Fertility & Early Pregnancy Working Group was approached by members of the ICC and NCS Program Office Staff and asked to prepare a formal response to the "Final Report from the National Children's Study Sampling Design Workshop" that was held on May 8-9, 2004 in Arlington, Virginia. On behalf of the Working Group, Dr. Germaine Buck presented a short talk to the Expert Panel underscoring the rationale and feasibility of preconception enrollment within the National Children's Study.

Members of the Fertility & Early Pregnancy Working Group were electronically sent a copy of the Expert Panel's Report in mid-May. Subsequently, the report was an agenda item for the Working Group's conference call on June 1, 2004. At that time, the Group agreed to offer a succinct formal response to the NCS as articulated below.

Members of the Fertility & Early Pregnancy Working Group applaud the efforts of the Expert Panel and strongly support their nine recommendations, though some are more directly relevant than others to the issue of preconception enrollment as noted below.

1. National probability sample is the preferred sampling approach. The Working Group strongly agrees that representation is important as further specified in the Expert Report. Moreover, the Working Group regards preconception recruitment as feasible with representative sampling as discussed in the collection of papers published by the Group. [Expanding Methodologies for Day Specific Probabilities of Conception, a workshop hosted by the Fertility & Early Pregnancy Working Group, provided further input regarding the methodology for preconception enrollment and the minimal data set required.]

2. Proportions of sample determined by different recruitment methods. The Working Group agrees that there are no apparent benefits to selecting study participants via different recruitment approaches such as selecting pregnant women and selecting couples preconception via another mechanism.
3. Standardized NCS protocol. The Working Group strongly agrees that the methodology for the NCS will need to be standardized including the centralization of resources, and that such approaches have been successfully implemented in other national studies as noted in the report.
4. Community and research buy-in. This issue has not been a part of the charge to the Fertility & Early Pregnancy Working Group, although we agree with the Expert Panel's observations.
5. Human reproduction and development encompasses biomedical and social factors. The Working Group recognizes that human reproduction and development involves interplay between biomedical, environmental and social factors at both the population and individual level. To this end, the Working Group strongly supports this point.
6. Capture of all pregnancies. The Working Group strongly supports the premise that post-implantation pregnancy (as determined by hCG) is the minimum unit of analysis for the NCS. Ignoring the loss of two-thirds of all pregnancies occurring among NCS participants will result in an irreplaceable loss of scientific knowledge, which is directly relevant to all aspects of the NCS. The technology for such capture exists and is likely to continue to improve in the near future. This approach is not inconsistent with the manner in which many women now recognize pregnancy (i.e., use of home pregnancy tests).
7. Inclusion of women at risk for pregnancy. The Working Group recognizes the potential for bias with regard to including only couples *planning* a pregnancy (though we are unaware of any empirical evidence that chemical or environmental profiles vary with regard to planning status) and supports the selection of households/women at risk for pregnancy to address this potential bias. The Working Group further recognizes the immense subjectivity with respect to planning, and the variability and imprecision of current approaches to measuring planning status in clinical or population-based research. Inclusion of women at risk for pregnancy underscores the role of behavior and other social factors that may impact a couple's decision

to actively attempt pregnancy or to allow pregnancy to happen. The Working Group emphasizes the importance of this point for all reproductive and developmental outcomes and not just rare disorders such as birth defects noted by the Panel. In addition, to the extent that severe disorders lead to early pregnancy loss, such disorders may be more common than anticipated.

8. Overrepresentation of geographical areas. The Working Group supports the need to ensure representation of individuals from geographical areas containing a wide variety of ethnic groups and high levels of exposures of interest. Various national databases exist and could be helpful in devising such a plan.
9. Streamlined approach for moving NCS forward. This point was not a part of the Working Group's charge, although the Expert Panel echoes many of our Working Group's concerns in this regard. To this end, the Working Group strongly supports conducting a purposeful and timely pilot study to refine methods for effective preconception enrollment.

In summary, the Fertility & Early Pregnancy Working Group supports unequivocally the points enumerated in the Expert Panel's report. This includes the need to obtain a nationally-representative probability sample, the inclusion of couples at risk for pregnancy, the inclusion of a range of environmental exposures, the inclusion of a range of reproductive & developmental outcomes, and recognition of the importance of social and behavioral factors in the context of biological determinants of health processes. Such a study design is best suited to the discovery and understanding of agent specific critical windows for the full spectrum of reproductive and developmental outcomes of concern to human development. We believe that preconception enrollment is the unique and most distinct and promising aspect of the NCS and that offers many scientific advances beyond those available from the many currently ongoing prospective studies that have relied upon recruiting women with clinical pregnancies.

To: Dr. Mattison

From: The Community Outreach and Communication Working Group

Subject: Comments on Possible Sampling Designs

Date: June 21, 2004

Thank you for the opportunity to comment on the proposed sampling designs. Given the charge of our committee, we offer input on the various sampling designs in light of our many previous discussions on the role of community engagement throughout the study (an aspect that we feel is crucial for ensuring the quality of the study- For a discussion of the rationale for and role of community engagement in the NCS, please see our document entitled "Community Outreach and Communications Working Group Guidance Document") and not solely on the scientific merit of the various designs (although we do feel that community engagement can help with recruitment and retention which will help to improve the quality of the science of the study). The following are our thoughts about the various designs:

1. We believe that community engagement is not only advantageous but crucial for the success of the study and believe that the sampling design for the study will need to facilitate community engagement in the support of the study. As such, we believe the use of a probability design (in which widely dispersed arrays of randomly selected locations will be implemented) will make this type of community engagement much more difficult, if not impossible. If participants are dispersed among a wider area without a concentration of participants in a geographical area, the identification and involvement of community-based groups and key influential persons becomes much more difficult in terms of locating a nucleus of these groups and persons that are representative of and relevant to the participants.

2. Our discussions to date have assumed some type of center-based model that will allow a geographical concentration of participants. We believe that the medical center model due to its focus on a geographical area (and hence, a less widely dispersed population of participants) is the closest to the model we feel is necessary to facilitate community engagement since it will more readily allow for the involvement of influential community organizations and individuals in some type of advisory capacity to the study. Our group also feels strongly that a medical center based model would need to require the involvement of other entities in the conduct of the study such as community health centers and nurse-managed health centers since these organizations have a strong record of service to populations that may not be served by the medical centers. In addition, to assist in the recruitment of women not currently in the medical care system, community based organizations and agencies would also need to be involved.

3. We appreciate the point made in the Final Report of the Sampling Design Workshop that separation from the medical care system may offer advantages in marketing the study by freeing the study of any perceived negative aspects of medical research. However, we point out that the study will still be recognized as "research' among those who are asked to participate and assume that even in the probability sampling design, participants will be informed that this is a government sponsored study. Given the mistrust of government sponsored research in light of examples such as the Tuskegee study, we are not sure that moving out of the medical centers will solve the problem of mistrust of medical research.

In response to this issue of mistrust, our committee has spent considerable time brainstorming proposed "RFP criteria" that could be used in selecting organizations to undertake the study to ensure the applicants have the type of relationships and reputation to diminish some of the mistrust of the medical care system. We have suggested that the criteria to select applicants include the following: a) Applicants should be able to affirm and demonstrate a history of productive community engagement as "equal partners" as well as an existing collaborative relationship with a university; b)Initial applicants selected should be those with a track record of community participatory research and an existing information network; c)applicants should have a process in place to demonstrate a sustained plan to recruit a cadre of advisory board members and establish student and adult internships from the study community

These type of criteria seem to be more appropriate for a center-based design than a probability sample.

4. While we agree that the use of pilot studies may be useful in exploring methods of community engagement (and especially methods of communicating the study and resultant response rates), we note that the suggested pilot study around "fostering community commitment" outlined on p. 6 of the sampling design workshop report would not answer the most crucial question around the potential benefit of community engagement and commitment to the NCS- the sustained involvement of participants over the life of the study. In our Expert Panel Workshop of November 2002, the participating academics suggested, based on their experience, that community engagement could help to increase the sustained involvement of participants.

5. Also, if the NCS is to investigate aspects of the community social and physical environment that can impact on health, it would seem that a center-based model would allow for the geographical concentration needed to implement some of the innovative methods (neighborhood checklists, GIS mapping, etc.) that are beginning to be more widely recognized as measurement tools for assessing the social and physical environment.

We look forward to discussing these and other issues during our conference call with the NCSAC.

To: National Children's Study Federal Advisory Committee

From: Study Design Working Group

Date: June 24, 2004

Re: Final Report from the NCS Sampling Design Workshop

Introduction:

Attached are comments on the sampling panel report that have been received to date from 16 members of our working group. We have had one phone meeting (June 21) to attempt to synthesize these comments. From the outset we recognized, as have you, the impossibility of making final decisions on sampling design for the NCS, without decisions having been made about specific hypotheses to be assessed and data collection methods to be employed, results from formal white papers of the existing literature and pilot testing of the initial feasibility – at least in terms of enrollment. Moreover, the working group had too little time between its receipt of the Sampling Workshop Report on May 28th and the June 28-29 meeting of the FAC to make a complete assessment of the report or to have engaged in a full discussion of alternative sampling methods. That said, members of the SDWG recognize four alternative approaches to sampling for the study.

- Population-based sampling using households as a sampling frame
- Population-based sampling using prenatal care clinics as a sampling frame
- Center-based sampling
- A hybrid or mixed model involving population-based sampling (using households or clinics) and center-based follow-up

Consensus needs to be guided by empirical data – historical and new pilot data - on that is the best sampling approach to take. The SDWG has therefore elected to provide a discussion of some of the issues we have identified, most of which revolve around the question of feasibility. The following discussion should be read as an executive summary from our working group that is neutral on which model is "best", but which we hope addresses issues of feasibility in a way that can help you discriminate between several approaches.

Discussion:

As discussed in the final report of the NCS Study Sampling Design Workshop, the SDWG agrees that there are important feasibility issues that will need to be addressed independently of the final study design. The primary issues that were discussed by the SDWG members include:

- Representativeness - How representative will the study population have to be in order to address key hypotheses? Is the study population to reflect the general population or specific subpopulations? How will participation and attrition impact representativeness?
- Life-Stage of First Assessment – Given study hypotheses, at what life-stage will the first assessment (and therefore the recruitment) need to be taken? In other words, what proportion of the population is to be recruited preconception, in the first trimester of pregnancy, or later? How will these participants be identified and recruited in a timely fashion?
- Burden - What will be the burden on participants? What will be the burden on health care providers?
- Retention – What will be the target for participant retention, and how will this target be met? Retention seems particularly difficult during the first year after birth.
- Data collection – What data collection will be required to address the study hypotheses? Will the same data be collected for all participants or will a set of "core" data be collected for the entire population, with additional subsets of more intensive sampling for particular hypotheses?
- Measurements/biological samples required – What measurements or biological samples will be required in order to address study hypotheses, and what means are available to collect these samples in a standard, valid and consistent fashion?
- Data standardization – How will sample collection and measurements be standardized among multiple sampling locations? This is particularly challenging for data that is to be collected at the point of contact with medical providers, such as prenatal ultrasounds, cord bloods, and placentas (assuming such data collection is required to address study hypotheses) given our highly diverse medical care system.
- Data variability - Will the study tailor data collection procedures to unique language, cultural and environmental circumstances in local communities and, if so, how?

These issues, available experience, and the literature were considered within the context of each of the four study approaches.

Population-based household sample: The Sampling Design Workshop Report suggested that a population-based household sampling strategy might successfully recruit subjects, take periodic surveys, and collect biological specimens for children, youth and adolescents over a long period of time, with good subject retention. The SDWG identified three possible approaches that could be construed as population-based sampling:

(1) Recruit women of childbearing age in households, continue to follow them until they become pregnant, and then recruit pregnancies;
(2) Recruit women in households in the first trimester of pregnancy;

(3) Recruit second babies prenatally, using birth certificates of first babies as a sampling frame.

The first of these options is the method discussed by the Sampling Workshop Report. For any of these options, recruitment could be door-to-door or by telephone. The SDWG could not identify any examples from the literature of a household-sampled preconceptionally or prenatally initiated birth cohort study of substantial size, requiring biological specimens, which has used any of these approaches. It was agreed that it would be very difficult with this sampling framework to obtain data that are associated with medical encounters such as prenatal ultrasounds, placental tissue, and umbilical cord blood, because the prenatal providers and hospitals of birth will generally not be known before the participant is pregnant, and will represent a cross-section of providers and hospitals, many, presumably, without the special expertise needed by the NCS. Such a sampling plan would also not allow for prenatal providers and hospitals to follow common data collection protocols and use common forms. Pilot work could determine whether women might be willing to go to a NCS study center to delivery instead of being delivered in a location and by a provider of their own choosing. Indeed some of the proponents of this approach seem to suggest that the NCS should give up the idea of collecting such biological and clinical data or get select biologic specimens as was done in Denmark. It was agreed that marriage licenses would be a poor base for recruitment since around one third of babies are now delivered to unmarried women. Some additional difficulties with the preconceptional recruitment model were raised. The first is the problem of the large number of women who would have to be followed in certain age groups, such as teenagers or women over 35, in order to obtain a live birth. The second difficulty was whether there is any assurance that the participant will remember to report her pregnancy to the study office in a timely enough fashion so that pregnancy data can be collected, especially if that data is time-sensitive, e.g. required to be obtained in the first trimester.

Population-based prenatal clinic sample: At this time, first trimester prenatal care is received by about 85% of women who give birth to a child in the US. It was suggested that prenatal care services could serve as the sampling frame for this study. An EPA focus group study designed to inform planning for the NCS has recently reported that women would prefer to hear about the NCS if their physician introduces it to them.[1] It was noted, however, that it might be difficult to gain the cooperation of all prenatal care providers in a given area and that resources would surely be needed to defray the effort that would be required on the part of providers, given that they are already “stretched thin”. A clear advantage of this approach would be a reduction of burden on participants, who would already be attending clinic, as well as the feasibility of obtaining clinical measurements in this setting, which may increase recruitment and retention rates. A clear disadvantage of this approach is that it will miss certain pregnancy data and specimens on women who come late, or not at all to prenatal care. (Currently about 2% of pregnant women have no prenatal care; such women are

unlikely to be easy to follow in any study). A difficulty is that any individual prenatal clinic is likely to have selected patients. However, there may be ways to sample clinics so that in aggregate they produce a sample of women reasonably representative of women receiving prenatal care in the region of the study. As with the first option, it would be necessary, in this model, to incorporate hospitals of birth into the sampling frame, if birth specimens (placenta, cord blood) are of interest to the NCS. Pre-pregnancy recruitment is of course not feasible with this model, and if the study hypotheses require pre-pregnancy sampling, either household sampling or a plan to follow next births to identified pregnancies will have to be employed for at least part of the study population.

Center approach: One of our members (BE) provided to the group a summary of efforts by the five Children's Environmental Health Research Centers that was prepared for the NCS. It showed a relatively poor response among prenatally recruited subjects, and considerable loss to follow-up. Participant burden was high, and this may account for the problems. However, reasonable financial incentives ($50 per visit, $300 for full data) were used in the studies. Much attrition occurred in the first year after birth. One other study was cited (NP) that has been able to follow higher proportions of children, into adulthood, even in inner city populations, but enrollment occurred at school age. A clear advantage of this approach is the ability to collect clinical measurements from participants, without having to make arrangements with myriads of health professionals in hundreds of care facilities, as would be required in one form or another for the population-based approaches. Such a sample can assess exposure-disease relationships. Another advantage is the ability to focus on particular at-risk populations. However, a clear disadvantage is the inability to develop true population norms for child development or population statistics about the prevalence of various exposures.

Mixed approach: In some geographic areas it might be possible to combine approaches, for example, to recruit subjects from a population based sampling frame (households or prenatal clinics) and ultimately to follow them from an academic health center or university. This model would be most applicable in areas where a large proportion of regional prenatal care providers have delivery privileges at the participating academic health center or centers. Some SDWG members think that the NCSAC should carefully explore whether such a mixed or hybrid model might be able to best provide an initially representative sample (which would become less representative over time with attrition). Although this approach was not recommended in the final report of the NCS Study Sampling Design Workshop, from a pragmatic stand point this option needs to be considered further, as it may provide a practical and affordable design to address the broad scientific goals of the study.

Adolescents: Throughout this discussion it became obvious that it may not be feasible to enroll and retain adolescents in any protocol in which the participant is enrolled prior to conception. Recruiting through prenatal care would be the most

practical way to enroll teenagers Retention of a cohort of adolescent mothers would be difficult no matter what, and the sense of the discussion is that the NCS will have difficulties obtaining a representative sample of teen births Also it would be important to consider whether the pregnant adolescent is viewed as an "emancipated minor", a condition that varies by State, complicating the ability to carry out a single protocol on a national basis.

Recommendations:

No consensus on the sampling plan for the NCS was reached by the SDWG. Indeed there are members of the study design-working group who have "grave doubts" about the feasibility of any of these models for long-term follow-up, particularly if population-based and the participant burden is high. In other words, some group members doubt that the study is feasible, no matter what approach might be taken. At the other extreme is a view that the study is so important that a "leap of faith" should be taken and that the study should move forward. In between are views that would support pilot studies and feasibility studies, perhaps using vanguard centers, in order to work through the feasibility issues before moving forward with a large scale effort, or to use a more tried and true design that recruits within the medical care system but works hard to obtain representative populations. The feasibility of the various proposed sampling schemes can also be estimated via a careful review of the many existing studies which put demands on participants that are similar to those proposed for the NCS, since only those will mirror the retention issues likely to occur in the NCS. Whatever approach to sampling is taken, the NCS will need to take great pains to assure standardization and consistency in the data collection across all sites.

With more time, and particularly with more information about the final choice of study hypotheses, SDWG can provide more specific input in the future. While ensuring a high level of follow-up is extremely important to the success of the NCS, the final design must be optimized to ensure collection of core hypothesis related data. For example, if cord blood and placental specimens are required for a core hypothesis, the final design must optimize collection of these specimens. Ultimately, the ideal sampling plan cannot be determined until it is known precisely what hypotheses are to be examined and what data elements are to be collected.

The following members of the SDWG were unable to participate in the teleconference discussion that led to this memorandum: Trudy Berkowitz, Frank Furstenburg, Yonette Jones, John Kiely, John Lynch, Louise Masse, Greg Pavlov, Mervyn Susser, Ira Tager,

[1] Lobdell DT: Identifying recruitment and retention issues for the National Childrens Study. Presented at the annual meeting of the Society for Pediatric and Perinatal Epidemiologic Research, Salt Lake City, Utah, June 15, 2003

SUMMARY OF COMMENTS FROM SDWG
RE SAMPLING PLANS FOR NCS

June 24, 2004

COMMENTS ARRANGED IN ALPHABETICAL ORDER OF COMMENTATOR, WITH DATES OF ISSUE, AND NOTATIONS AS TO WHETHER THEY WERE RESPONDING TO A COMMENT FROM ANOTHER MEMBER OF THE SDWG

FROM TYE ARBUCKLE 6-17

My first response to the recommendation by NSAC for a national probability sample was oh no, the survey methodologists are leading this, especially when I saw one of the contractor's reports talking about sampling weights. However, after reading the sampling panel report, I began to see some advantages to the probability sample versus the medical center approach and questioned my concern (and those of others in opposition to this approach that retention would be more difficult). The characteristics of a geographic region- based national probability sample with oversampling of specific socio-economic/ethnic groups that changed my opinion are:
a) opportunity for a more complete representation of the community, a community-based rather than center-based study might be easier to "sell";
b) an unbiased preconception cohort and no need for a separate sampling approach to include this segment;
c) collection of "exposure" data on pre-conception women AND men;
d) collection of "outcome" data on infertility, spontaneous abortions, etc;
e) some concerns about whether a medical center approach would miss segments of the population not entering the "system" (e.g., home births) or entering the system later in pregnancy;
f) focus on the woman or couple rather than on the participating medical center to provide information, specimens and consent to access medical records;
g) at least at the start of the study, an indication of how representative the population and pregnancies are - for example, can compare births with birth records; I agree that at the end of the study, the study population is unlikely to be "nationally representative";
h) ability to generate attributable fractions;
i) sampling unit is not the pregnancy but the couple and all their pregnancies (prior & subsequent) - good for gene-gene and gene-environment studies;
j) the same sampling approach could be used at all sites;

As I expressed earlier, my main concern with this approach was feasibility. I agree with the panel that there needs to be focus group and pilot work done with both approaches to assess the feasibility of either approach. For the probability sample, during recruitment, the subject could be asked to identify their medical care provider and likely hospital to determine how many providers would have to be approached for IRB and access to medical records and specimens. I have not heard this discussed, but is there any opportunity for using parts of the NHANES survey (i.e., reproductive age men and women [and their respective spouses]) to identify and recruit the population for the NCS? You could take advantage of all the work already done for this survey (e.g., mobile units, infastructure, standardized questionnaires, biological samples and physical measurements). Recruitment could come from previous NHANES

participants plus current and future participants. I recall one of the first presentations that I saw on the NCS - THINK BOLDLY!!

TRUDY BERKOWITZ 6-14 (RE LYNN GOLDMAN'S COMMENT OF 6-10)

I think Lynn's third option represents an important contribution to the discussion of the optimal sampling strategy. It would incorporate some of the scientific advantages of probability samples with the wealth of experience and expertise by the various academic centers that have been in the business of carrying out longitudinal pregnancy studies.

FROM JONAS ELLENBERG 6-9

I was able to sit in on the first day of the deliberations of the workshop panel, as a member of the planning group for the workshop, representing the SDWG. The panel interacted very constructively, asked probing questions, and, in my view, provided a consensus that was appropriate to the evidence presented at the meeting or generally available in the public and private domains. My read is that the panel made a very simple series of findings. A national probability sample is the most scientifically appropriate design for the NCS; there is a need to show in pilot work that such a design is feasible. In determining these major findings, they also made a finding that there is no credible evidence that either accrual or retention will be more or less difficult for either the national probability design or the center-based design. Thus, their final finding is that the probability design is appropriate and that pilot testing of both the national probability design and the center-based design for proof of feasibility (the center-based design as back-up) is an appropriate next step. I concur with their logic and their findings. I look forward to discussing this further on our conference call on the 21st.

FROM BRENDA ESKENAZI 6-10 (RE ELAIN HUBAL'S COMMENT OF 6-10)

I can report on the results of a recent paper summarizing the experiences of the Centers of Children's Environmental Health Research and Disease Prevention in which both Trudi and I participated. Five of the Centers are conducting birth cohort studies of the level of intensity that has been discussed by the NCS albeit much smaller. We have been asked to summarize this by the NCS and these papers are currently under review by them and will be published as a supplement in EHP. Our experience would strongly suggest that a probability sample would not have adequate follow-up and that those that will agree to participate over the long haul will not be representative. Thus, I believe a probability sample may work for a less intensive study but not for a study which requires follow-up for 21 years with intensive biologic and

environmental sampling --In this case, a center-based approach may be the only feasible one.

SECOND COMMENT 6-20

Five Centers are conducting birth cohort studies (N~500). We recruited diverse populations, including low-income and various race/ethnic groups (Appalachian, Dominican, Hmong, Laotian, Mexican, and Puerto Rican). Our Centers worked closely with our respective communities to develop partnerships, strengthen community infrastructure, build trust, and conduct more culturally appropriate research.

Response rates for the studies ranged from 25 to 60 percent. The most important barrier to participation was the time required for each individual visit as well as the length of the follow-up period, especially for working women. Centers that recruited patients from the clinic waiting areas found that short waiting periods, especially in private practice offices, were a barrier. The one Center that used clinic staff for recruitment found that these staff were already overburdened and had little time for recruitment. Some Centers also found that women were reluctant to enroll without their husband's approval.

The loss to follow-up rate for Centers that have completed the two-year visit ranged from 15% to 26%. However, because some Centers did not include participants for follow-up who did not complete certain pregnancy events or the child was considered to be at high risk, it is difficult to compare retention rates across Centers. The greatest losses occurred during the prenatal period and before the child was 12 months. Most Centers found that their study populations stabilized once the child turned one year old, and some Centers had better response rates at 24 months.

These Centers have allocated about 500K per year of study (total Center allocation for direct costs~1 million/year); therefore, from **pregnancy to age 2 years cost about 2.5 million (including startup time). The average N=500. Although there may be some savings for a larger sample, this would translate into a cost of about 500 million for the first five years for an N=100,000.**

FROM FRANK FURSTENBURG

FIRST COMMENT 6-11

From afar, Italy, I've been reading and thinking about the issues raised by this discussion. As someone who has carried out a series of longitudinal studies, some based on probability samples and some on purposive samples, I strongly agree with the general consensus that it if far better to begin with a probability sample. The benefits of the data set, of course, diminish with attrition, but there are sound techniques for dealing with attrition (imputation of missing

values, weighting, as well as ways of strategically re-sampling missing cases in later waves) that help detect biases created by attrition.
Moreover, clinically based samples are not free from attrition. The problems of attrition may be created by mobility (more easily attacked especially when tracing information is abundant and resources are great) than burnout. But there are also ways of dealing with burn out. Promising less frequent follow up, incentives, and skilled interviewers who are capable of maintaining ties. Newsletters with relevant findings and cards help a lot. But the point is that for many issues of relevance to social scientists, clinical samples simply don't do the job, and as others have pointed out, may be misleading. Think, for example, of the problem of exploring differences among new immigrant populations or social class differences which may influence the impact of treatment and outcomes. For medical purposes, such purposes may be irrelevant, but for social polity regarding exposure and access, they could be enormous.

SECOND COMMENT 6-19

There are many long-term follow up studies of nationally representative samples that have had reasonable rates of retention in addition to NLSY. To mention but a few, National Educational Longitudinal Survey (NELS), Panel on Income Dynamics (PSID, ADHealth, National Survey on Families and Households. All of these studies have ranged over many years, many including multiple family members, and most have achieved response rates that are as good as many clinically based samples. Time and money are the chief predictors of response rates that reach above the 70 percent range after many years and often go much higher. The NYSY has included clinical assessments of children and the PSID has asked parents for time diaries. The list goes on.

FROM LYNN GOLDMAN 6-10 (RE BRENDA ESKENAZI'S COMMENT OF 6-10)

I agree with Brenda. At the end of the day it won't be a probability based sample any more, because of the fact that those who participate in follow-up studies over 21 years will not, by definition, be a representative sample. However, I also agree with Elaine and others, that there are large questions that an initial probability sample can address. Initially at least it should be possible to recruit a pretty good probability sample. I wonder about a third option. I am thinking about the study design that is being utilized by Southampton Women's Survey. Women 20-34 years old are recruited on a population basis and baseline interviews are taken. Those who become pregnant are invited to take part in the pregnancy phase of the survey; researchers at the University of Southamptom do ultrasound scans at 11, 19 and 34 weeks of pregnancy, and then babies are studied at birth, and ages six

months, one year, two years and three years. It seems to me that the NCS could take an approach that would utilized a probability based sample of PSUs across the US and one or more contractors to do recruitment and initial data collection on women. Academic centers (perhaps in conjunction with contractors) could then carry out follow-up investigations. Such investigations could include data collection standardized across the nation (i.e. for questions requiring larger numbers) and other data that might be collected for smaller populations and possibly more directly relevant to the research interests of academic centers. Would it not be possible to find a way to design this study that would provide both the benefits of a probability sample and engagement of Centers, to assure adequate follow-up? Administratively, I do know that there is a kind of federal agreement called a cooperative agreement that allows the govt to do work that is a hybrid between a grant and a contract, so I think that this kind of approach to study design is feasible in that respect.

FROM ELAINE HUBAL 6-10

I believe that the questions that we are trying to address under the NCS are significantly different than those that have been addressed historically using the center-based model. With NCS we hope to improve our understanding of the relationships between environmental exposures and health outcomes for our children as a function of genetic, behavioral, and community factors. The impact of the study will be to improve the scientific basis of public health and environmental policy decisions. Therefore, I strongly agree with the conclusions of the workshop panel. A national probability sample is the most scientifically appropriate design for the NCS. And there is a need to show in pilot work that such a design is feasible. Should preliminary pilot work indicate that the national probability sample model is not feasible, then we will have to settle for the center-based model and make it work. As such, pilot work to address extending the center-based model to recruit a more representative sample should also be conducted.

FROM MARK KLEBANOFF 6-18

The NCHS did, as I recall, conduct a follow-up some time in the 1980s of the individuals samples in the early-mid 1970s as part of the NHANES study to determine mortality. I think they also administered a questionnaire. They may also have followed those people again more recently, but I can't recall. The National Longitudinal Survey of Youth has followed a probability based sample of people who were adolescents/young adults in around 1979, I believe. They are still following them today. The main goals of that survey, as I recall, related to the experience of the cohort in the labor force, although they have collected health information on the cohort, as well as information on their pregnancies and any children they might have had. I gather that tracing and

contact has been fairly successful over the years. However, as far as I can recall, all their contact has been questionnaire based, either by phone or mail. There are no exams, nor any biological specimens-- at least that I recall. Neither I, nor as far as I know any colleague in reproductive, perinatal or child health epidemiology, has a very high opinion of the quality of the self-reported, unverified data collected relevant to our area; and it's rare (or maybe non-existent) for reports on these topics from the NLSY to make it into a first-line medical or epidemiological journal. The quality of the data on the economic condition, etc. of this cohort may be fine-- I am not qualified to judge. Those are the only studies that I'm aware of that might even have a chance of shedding light on this question.

FROM ROD LITTLE

FIRST COMMENT 6-10 (RE IRA TAGER'S COMMENT OF 6-10)

Oversampling for environmental exposures is just as feasible in the probability sampling approach as in the medical center approach, indeed I would argue that it is easier to formally incorporate this into the probability sampling approach; so to my mind this is not an argument for the medical center model. I have argued that this and other forms of oversampling needs careful consideration, and I think it should be a important topic for the next FAC. No one is arguing that having strong scientific hypotheses is crucial to the study, and whatever the results, no one can doubt the study planners' unprecedented efforts to develop them. As a statistician I'd say study design also plays a crucial role -- a poorly designed study addressing a question of high scientific importance is still a poor study; the selection of subjects affects every single hypotheses addressed in this study to some degree, and hence for me it also has a high scientific importance. I'd like to commend the sampling committee for their fine report.

SECOND COMMENT 6-16 (RE MERVYN SUSSER'S COMMENTS OF 6/11)

It is true that limiting attrition is a key within any model; I would argue that a centralized approach to retention (as in a probability sampling model) is more promising than an approach that leaves retention efforts to a set of centers (particularly if that set is large). With multisite studies it has been my experience that efforts and results tend to be pretty variable across centers. Also there are major issues with migration between areas given the long period of the study. I think it is scientifically misleading to use overall participation and retention rate to compare across probability and center-based sampling models. A volunteer-based sample will likely look better on these measures, but that is because a key aspect of the probability sampling model is the attempt to include people who would not volunteer, and I think the goal is to make

inferences about a population that includes non-volunteers as well as volunteers. For example, health services folks may have important questions about the impact of access to health care, and a sample of volunteers to medical centers may be a very distorted sample for addressing such issues, since it may miss individuals dissatisfied with the health system.

The goal of retaining a probability sample of the population of volunteers and non-volunteers is not attainable, but it seems to me clear that one can get closer starting with a probability sample than starting with a volunteer sample. In that latter model, the probability of inclusion of non-volunteers is zero; in the former, the probability of inclusion of non-volunteers is probably lower than that of volunteers, but it is not zero; and nonresponse weighting strategies may reduce the bias from relative under-representation of the non-volunteer population. Having something is to my mind much better than having nothing. Another point is that it may be possible to measure some things on hard-to-retain individuals but not others; it is better to have partial information on them than to have nothing.

Clearly my division of the population into "volunteers" and "nonvolunteers" is a simplification, but is intended to make the issue clearer. Note that taking a probability sample of psu's (which clearly I strongly favor) but then fudging the second-stage selection by having centers do something like a quota or convenience sample within demographic groups does not result in a probability sample overall, and is subject to the biases of a volunteer sample noted above. One could have centers attempt to collect a probability sample at the second stage, but I wonder if they are best equipped to do that --- I think it would be good if the rfp process was strict enough to limit the study to centers that would be so equipped, but I question whether it is the optimal solution.

I do think that academic centers should play an important role in the study, I just think that sampling and retention issues are best centralized. Finding the right organizational structure for the study seems to me a key. Several of us, including myself in my first response (that seems to have disappeared? perhaps I failed to hit "reply all") suggested the need for pilot studies. But a pilot could hardly solve some of the critical unknowns. Can a pilot of relatively brief duration tell us about what is crucial, namely, the likely participation and attrition rates in a longterm national probability sample over two decades? A necessarily brief pilot seems unlikely to predict losses over so long a period. Here we are sailing into unknown seas.

I know Peter is not interested in a pilot study that attempts to settle the retention rate issue, and I agree with him. Pilots could certainly be useful in establishing practical aspects of different designs. Participation rates might better be guessed by drawing from assembled experience of previous studies;

even so, we must allow for changes over time since they were undertaken. But attrition rate is the most critical datum. Perhaps one might reasonably extrapolate from the experience of losses over time in the (meagre?) assembled literature bearing on all relatively large longterm studies beginning at birth or soon after? Has anyone put together such material? Those data might yield at least a reasonable guesstimate of participation and loss rates within the earlier years of life (a decade at least in the NCCP, I think), especially if the data could be re-ordered in classes and proportions that resemble the expected distributions in a national sample. My sense is that the Battelle group tried to do this, but left out some key examples. Anyway, as noted I think this is a key issue with any design, and not a basis of deciding between them. In the end, this enterprise may come down to mustering the courage to sail into the unknown, with only the hope that material and other costs will be justified by the results. Of course, as others have also stressed, it is essential to frame the whole undertaking in terms of what the hypotheses demand if they are to be adequately tested. But we can't brush aside the difficulties (nor the discomfiting thought of the billions at stake). Many a brave sailor, Captain Cook among them, perished on voyages into the unknown. Anchors Away!

THIRD COMMENT 6-20

1. I liked the expert panel report a lot, and my impression is that a lot of other people did to. The report was very clear and concise, and clearly gave a boost for the probability sampling model, though both models were addressed. I also liked the emphasis on what to design into pilots, and think that should be a primary focus of the next FAC.

2. I believe that participation of "academic medical centers" is to be in the list of "required characteristics of the design", which underscores a perceived need for this that was previously implicit. I think that a relatively centralized organization overseeing the sampling and fieldwork has strong advantages, and does not preclude involvement of the academic community (as for example is done through the PSID Board of Overseers).
Key issues I see here are

2a. Who should do what in this study, to achieve the scientific aims? In particular, given that academic medical centers should play a key scientific role, what is the appropriate way of involving them? If probability sampling and retention are viewed as key ingredients, how is expertise on these aspects built into the study?

2b. Perhaps most crucial, what is the best organizational structure for such a vast study? This needs careful thought, and the FAC could provide useful input (as could the design working group).

3. I think we need to get beyond discussions of the different models -- probability, center, combinations -- with pros and cons. My opinion is that the two expert advisory groups that have been asked have both opted for a probability design to the extent feasible, and it is time to face up to the reality of trying to make that model work. I think some like the idea of giving the study to the medical centers but designing in probabilistic aspects. It's worth exploring but am not sure that probability sampling and recruitment is what medical centers do well, and a more a centralized organizational structure is needed.

4. Peter's group has been working with Randy Curtin on sampling design aspects -- number of clusters, definition of the ultimate clusters, feasibility of getting individuals in these clusters to medical centers for medical measures, etc. I think this is good. I am a bit concerned that the compromise may be a design that starts with a probability sample of PSU's, but then fudges the probability selection of second stage units. I think it is important to keep in mind that if the selection at the second stage is of volunteers, then this is not "representative" in any real scientific sense – no one has successfully defined "representative" outside a probability sampling framework, and response rates aside, the key limitation of volunteer samples is that non-volunteers are not represented.

5.Too many people still equate "probability sample" with "equal probability sample", and I'd like to reiterate that oversampling of certain groups or locations is readily accomplished within the probability sampling model. Whether such oversampling should be undertaken is a very important question. An equal probability design becomes attractive as the aims of the study broaden, but I have thought from the outset that some oversampling of environmentally contaminated sites is worth serious consideration. Of course the question then is what specifically do you mean by contaminated, but I think some kind of "bad stuff" stratification could be developed. This would of course correlate with "poverty" and "race" to some degree, which would please some groups, and I prefer a stratification more explicitly tied to scientific aim of impact of environmental contaminants on disease.

6. Another key design issue concerns the early conception piece. Some believe that a broad screen of potential pregnant women to achieve the full sample is much too expensive. The question is what can be done to do this more economically. It's a good question.

7. I also think an important issue is what pilots can be conducted to elucidate major design issues, without seriously delaying the study; there may be a trade-off between the need to maintain the existing time-line to keep people interested, and the need for pilots that increase the chances of success of the study.

FROM JOHN LYNCH

FIRST COMMENT 6-15 (RE LYNN GOLDMAN'S COMMENT OF 6-10)

I tend to agree with Lynn's suggestion for the hybrid design that was also discussed in the Battelle report. The national probability sample has scientific advantages for recruitment - if indeed it is feasible - and Nigel has laid out some of the important questions that will need to be answered in that regard. I am also concerned about retention and as Jonas reported, while the sampling group found no credible evidence for the superiority of either design, I agree with Mervyn that we are in uncharted territory here in regard to length of follow-up and intensity of proposed data collection and have to just use our best judgement - there is no pilot study that can tell us which design will be best for retention. I can't help feeling that this study will have to make a very strong connection with these mothers, kids and families, especially given the large subject burden associated with the diverse types of data collection being proposed. I recently sat on a review panel for the ALSPAC study in the UK and the ALSPAC families are getting questionnaires every quarter in addition to clinic visits and the scope of data collection for the NCS is likely to be even broader than ALSPAC. This only gets worse as the kids get older and have their own agendas for how to spend their time. Its going to require lots more than newsletters to keep the NCS participants engaged - they will have to connect with a place that has continuity of staff and there will have to be lots of community-based mobilization of advertizing, special events for the families, annual picnics, school visits, regular local news coverage, and local corporate support all happening over an extended period of time - these communities and participants will have to feel some sense of ownership of this study and its benefits to them will need to be tangible and meaningful. I don't know how to achieve this or exactly what implications this has for either design but the national sample approach seems more amorphous to me and may lack the clear identity that comes with the centre-based model but perhaps I don't have the right vision of it. Finally, I think the attrition will unfortunately be large in whatever approach is used and while imputation can help, it can't solve the problem of large amounts of missing data so maybe we may need to think about staging the study and reconsider replenishment samples at important age transitions.

SECOND COMMENT 6-19

My impression is that the British birth cohorts have much less extensive follow-up at much larger time spacing than is being proposed for NCS. Additionally, it has been primarily (in some cases exclusively) questionnaire until recently. Again NHANES follow-up was once or at best 2 times.The NLSY was based on pop sample of women to gain their labor market experience. From this a sample of kids was recruited as Mark says, but I believe there was reasonably high non-response and so the sample of kids is unlikely to be representative and again follow-up much less intense with no biological samples. All this can be readily confirmed.

FROM NIGEL PANETH 6-20

Our most critical need is to know more about the feasibility of implementing a population-based sampling frame involving non-pregnant women, but which seeks to ascertain key events in pregnancy and at birth. The experiences cited in sample surveys and other forms of follow-up of samples obtained from the general population have not dealt with the time constraints imposed by pregnancy and birth. Three particular difficulties must be addressed.

1. THE PROBLEM OF IDENTIFYING PREGNANCIES IN PARTICIPANTS. While the NCS is interested principally in enrolling pregnant women who will produce children, the population sampling plan recruits women of childbearing age. Thus the study is dependent upon:

   a. Identifying women who have a real probability of getting pregnant and

   b. Ensuring that these women report their pregnancies in a timely fashion to the study office when they occur

a. As to the probabilities of getting pregnant in a given year by age, please see the table below (sent on June 9 to the SDWG and to the NCS program office). For women at the peak of childbearing, age 20-35, one needs to follow some 8-10 women to achieve, on average, one pregnancy in a year. For women over 35, the figure is one pregnancy per 23 women, and for women in their early forties one in 120. If first births are required (as would be implied by an interest in primary infertility) one needs to follow more than 100 women per year in their late thirties, and more than 500 per year in their early forties. It is probably for this reason that the only study I know that attempted this, by Keith Godfrey in Southampton, UK (not yet published), restricted their sampling to women 20-35. Would we do the same in NCS? If not, what is a reasonable estimate of the expense needed to find first births among teenagers or among

women in their forties? Or would such populations be left out of the study? And if they are left out, in what sense is the study representative? In the Southampton study, 13,500 women were recruited, and this yielded about 400-500 pregnancies a year. Fertility is slightly higher in the US, but one must contemplate following at least one million women in their peak reproductive years to ascertain 100,000 pregnancies that will lead to births, and this plan would exclude women < 20 and > 35. I can imagine no economically feasible plan to follow populations whose annual fertility is less than 5% and hope to ascertain their pregnancies without considerable loss.

TABLE: BIRTH RATES AND FIRST BIRTH RATES TO US WOMEN BY AGE (N OF WOMEN FROM 2000 CENSUS; N OF BIRTHS AND BIRTH RATES FROM 2002 NATALITY SURVEY)

1	2	3	4	5	6	7
AGE	N OF WOMEN	BIRTH RATE per 1,000	FIRST BIRTH RATE	N OF BIRTHS	RATIO OF WOMEN TO BIRTHS	RATIO OF WOMEN TO FIRST BIRTHS
15-17	5,835,448	23.2	20.8	135,382	43.1	48.1
18 -19	3,993,438	72.8	54.1	290,722	13.7	18.5
20-24	9,276,187	103.6	48.1	961,013	9.6	20.8
25-29	9,582,576	113.6	40.7	1,088,580	8.8	24.6
30-34	10,188,619	91.5	26.6	932,258	10.9	37.6
35-39	11,387,968	41.4	9.3	471,461	22.7	107.5
40-44	11,312,761	8.3	1.8	93,896	120.5	555.6
15-44	61,576,997	64.5		3,973,312	15.5	

Column 6 above is a reasonable estimate of the chances of a woman of a given age having a live birth within one year, and column 7 of any woman having a first birth within a year. Pregnancy rates are of course higher (in some groups perhaps as much as twice as high). One has to consider that the numbers in columns 6 and 7 are a slight overstatement of the chances of encountering a woman liable to a pregnancy, since there is some probability that one will encounter a woman already pregnant at time of contact. Also, while these figures are annual, they are not cumulative, as the total (i.e. lifetime) fertility rate to women in the US in 2002 was just 2.013.

One can see that a population-based design that focuses on women 20-34 would require only about 8-10 women to be followed for a year to get a live

birth (assuming zero attrition, which is unrealistic), but for the other age groups the effort would be much larger. If one wants to target nulliparous women, e.g. to study infertility, the N of women to contact is about doubled except in the very youngest, and becomes perhaps prohibitive above age 35, where more than one hundred women must be followed to obtain one first birth in a year.

b. In Southampton, Godfrey reports that about ½ of the pregnancies in the identified women were reported by the women to the study office. The remainder were ascertained by NHS doctors who were paid to provide notification. We of course have no such medical system here. How likely is it that we will be able to keep in sufficiently close contact with pregnant women in the US so that they will notify the study in sufficient time to obtain the required pregnancy information?

2. THE PROBLEM OF OBTAINING BIOLOGICAL SPECIMENS DURING A SPECIFIC TIME WINDOW.

Even if women report their pregnancies to us, we may still have the problem of obtaining information (be it self report, measurement or biological specimen) during a narrow time window. If we want to obtain information in the first trimester, the largest possible window is 12-13 weeks, but this will be shortened by however long it takes women to establish that they are pregnant and report it to the study office. How would the study office arrange data collection in the short time period between a woman recognizing her pregnancy, reporting it to the study office and the end of the first trimester? Alternatively, are all hypotheses requiring time-dependent information in pregnancy to be discarded?

3. THE PROBLEM OF INTERACTING WITH THE MEDICAL CARE SYSTEM

Much of the discussion about population sampling derives from experiences in which all or most of the study information is obtained from the participant's responses. In some of the cited population-based studies, biological specimens are limited to serum and urine that can be obtained at any time in the life cycle. But the NCS has been considering obtaining more complex biological specimens in pregnancy, and these might have to be obtained at specific times of pregnancy, and at birth. The current NCS small business announcement speaks of 3-D ultrasound and other pregnancy technologies in the capacity statement, implying that some variables will require this kind of assessment. Is portable US equipment brought to the participant's home feasible?

If not, how are such studies to be done during pregnancy without contact with the prenatal care provider? How will the study office learn who the provider is

in timely fashion? Assuming we learn the name of the provider from the participant in timely fashion, how do we obtain their participation and ensure they have the requisite equipment? How many providers would we have to contact for every 1,000 sampled pregnancies? How do we obtain prenatal medical records without prior agreement with the provider, who will rarely be known at the time the women is entered into the study? What about the extreme variability in prenatal data collection across practices in the US? Is there any feasible way to standardize such data collection (as was done in the NCPP) without having an understanding in advance with prenatal care providers?

If the prenatal care provider is one difficulty, the hospital of birth is a second. For any 1,000 sampled women, how many hospitals of delivery would be involved? How do we get their participation? How many can reliably collect and store placentas, obtain cord blood specimens around the clock, provide protocol neonatal examinations? My obstetrical colleagues think that about 20% of US hospitals could obtain a cord blood specimen with reliability around the clock.

Below is a list of steps at which the population-sampling model is likely to incur data loss, to which all of us can attach estimates.

1. Initial refusal to participate
2. Woman participates, but is lost at some interval between enrollment and getting pregnant
3. Gets pregnant, but does not remember to notify study office
4. Notifies study office, but not in time to obtain specimens
5. Notifies study office in time, but prenatal provider is not cooperative
6. Prenatal provider is cooperative, but does not have requisite data collection capacity
7. Prenatal care provider has capacity, but hospital of birth not cooperative
8. Hospital cooperative, but does not have capacity to obtain specimens

My estimate is that starting from obtaining the population-based sampling frame, one would be fortunate to obtain 20% of women for whom all requisite data is obtained within the appropriate time window or some reasonable approximation thereof.

Now contrast this with the fact that 98% of women in the US visit a prenatal care provider, and 85% of them do so in the first trimester. Add this to the finding, just reported at SPER, of the EPA pilot study, which found in focus groups that women overwhelmingly preferred to be informed of the NCS by their health care providers**. Surely it is much more sensible and efficient**

to sample prenatal patients. While in any single venue they are a selected sample, it is possible to find collections of prenatal care venues that reflect the population of pregnant women in a region. Most of the difficulties listed above (getting the pregnancy reported, being able to obtain timely specimens, links to the medical care system) are not issues in this design. Moreover, a common prenatal data collection system for providers can be used, which is not feasible in the population of women sampling model. It will also be important to ensure that hospital delivery systems are also incorporated in the design, but again, there are regions in the US with a common set of prenatal and delivery care providers, which can provide reasonably representative populations of the US (testable through analyses of birth certificate files).

Since, as I note above, obtaining first pregnancies in a representative sample of some populations (e.g. > 40) is essentially not feasible, I would recommend studying preconceptional issues in the NCS by following primiparous participants until their next pregnancy.

PAUL SORLIE 6-17

Since a longitudinal cohort study requires successful participation and follow-up in future years, I think the arguments toward a sampling design should, in balance, stress successful follow-up over a strictly representative sample which would yield an excellent cross-sectional study. From all of our experiences in longitudinal cohort studies in the cardiovascular arena, we are convinced that successful follow-up, contact and re-examination of participants requires community involvement. It has been essential for all of our studies, including the older established studies such as Framingham. Our cohort studies in Native Americans, African Americans and consultations regarding studies in Hispanic Americans, all emphasize that if people are to commit their time, they need to see the support of the community, the medical institutions around them, and a return to them from the study, rather than the study only taking from them. I have attached a file with two tables showing participation rates in one of our longitudinal studies of young adults (CARDIA). This study began in the mid 1980's. These are participation rates in a repeated examination (requires attending a clinic exam) and participation rates regarding telephone contact. These tables are for women ages 18-30 at entry, and the study was cardiovascular, not regarding maternal or childhood issues. These participation rates require extensive effort as described above. If the sampling design does not permit this kind of continual support over the length of the study from community organizations, physicians, clergy, medical centers, etc, the response will be much lower.

Table 1: Participation Rates (%) at Each Clinic Examination CARDIA Study – a longitudinal cohort study of cardiovascular risk factors. Women, Age 18-30 years at baseline

Race/ Education	Number at Baseline	Year2	Year5	Year7	Year10	Year15
Black ≤ high school	703	85	80	74	75	67
Black > high school	777	90	85	81	78	73
White ≤ high school	354	92	89	83	77	73
White > high school	953	95	91	86	84	82

Table 2: Percent of participants alive who were successfully contacted by telephone 16 years after baseline; women, 18-30 years at baseline; (HS, high school).

Black				White			
Age 24 ≤yrs		Age ≥25 years		Age ≤24yrs		Age ≥25 yrs	
≤ HS	> HS	≤ HS	> HS	≤ HS	> HS	≤ HS	> HS
79	81	80	83	86	91	91	93

FROM FRANK SPEIZER 6-8

The sampling committee did an excellent job in answering the question posed to them, but they operated under certain constraints. They started with the premise that is indicated in the overview of the White Paper that "..the main objective of the NCS is to study relationships between exposures, including chemical, physical, biological, and psychosocial exposures, and outcomes." and that the NACS is to primarily an 'analytical' study rather than an 'enumerative' study. This seems reasonable in that by the time the study is over (20 years) the descriptive nature of the population will be different. I am therefore suprised that they focussed so heavily on a National Probablity Sample as option one with minimal discussion of set aside funds for investigator-initiated components. Option 2, although reasonably discussed did not appear to be as enthusiastically supported, although they did not rule it out and rightfully suggested some pilot work be done. I was surprised that there was not more discussion of the mixed model.

One major concern with either model, that was not fully discussed, nor do they propose pilot work for, is the concern that I have had for some time that relates to whether this is really a study of environmental risk factors. I see no evidence

that we have focussed any samplying discussion on the numbers of people we need to have exposed to be able to assess exposures of interest. In fact, we have yet to have the discussion of what the exposure of interest are!! This harps back to the concern that we have not yet seen specific hypotheses and therfore cannot make the estimates needed. I would agree with the sampling committee that this study should be planned to answer questions that cannot be answered by more traditional approaches (certainly if we are going to justify $3 billion) and we haven't seen that yet. It seems to me that once we begin to see the specific hypotheses we may be able to rule in or rule out the specific design options simply of the basis of the feasibility of answering the questions. Perhaps under the best of designs (assumming good retention and follow up) we may not have enough at 100,000 and therefore that hypothesis will have to be explored in some other design or by some other study not related to the NCS. If this come up in enough of the hypotheses maybe the $3 billion would be better spent is some alternative fashion unrelated to the NCS.

This is an intersting issue as I start to think about the 6000 hospitals in the US. Do we know how many births occur in hospitals and how many in birthing centers not in hospitals? Birth is just one outcome, what happens after birth to children? What proportion have a contact with a hospital within the first 5 years of life? I suspect less than 10% but that number might be known from National Survey data. We are therefore going to be involved with health care encounters that are outside hospitals most of the time. (This means to me self (family) reporting and validation of reports in selected samples THAT CORRESPOND TO THE HYPOTHESES OF INTEREST. Again the need to have the hypotheses.

SECOND COMMENT 6-10 (RE LYNN'S GOLDMAN'S COMMENT OF 6-10)

This third option might very well get us somewhere. It seems to me it would make a lot of sense to use a two phase screening. This would likely get a more generalizable sample of (non pregnant) as well as pregnant (much smaller number) of households originally and then use the academic centers to contract for follow up. Could the initial screening all be done by mail questionnaire, with perhaps a cheek swab put away on a much larger sample and therefore a more significantly weighted sample by potentially important exposures? Frank

FROM MERVYN SUSSER

FIRST COMMENT 6-9

It is surely clear, given the array of questions raised, that pilot work will have to be done, although even then we would not have answers all the problematic open questions. Certainly more substantial testing and enquiry is needed on the national probability sample than on the center sample approach. can not agree

that each is equally credible at this stage. Certainly there is much experience of probability sampling of a national character in the NCS from the many surveys they have and continued to conduct. But cross-sectional surveys are simple and bear little relation to the problems arising in 20-year and possibly lifelong follow-up in longitudinal designs: of these we have some experience of various sites, as with the National Perinatal Collaborative study and many others. One can assert that the experience garnered indicates that the Center- based design is certainly feasible. So we should not set sail on a probability sample approach until the pilot work demonstrates feasibility especially in terms of acquisition and retention of participants. The same degree of restriction does not apply to center-based studies.

Frank pushes the question of in or absent adequate hypotheses. Certainly there is much to do there. Perhaps that could be approached by setting up small working groups to address, with greater intensity than the whole group can muster, the development of those so far favored by those of us (and other available experts) who are best versed in the territory of each area into a form that meets at least the standards required for most new NIH proposals.

SECOND COMMENT 6-11

The discussion on sampling so far has been interesting and useful. Clearly, a fundamental issue is the attrition and participation rate over long periods. Several of us, including myself, in my first response suggested the need for pilot studies. But a pilot could hardly solve some of the critical unknowns. Can a pilot of relatively brief duration tell us about what is crucial, namely, the likely participation and attrition rates in a long-term national probability sample over two decades? A necessarily brief pilot seems unlikely to predict losses over so long a period. Here we are sailing into unknown seas.

Participation rates might better be guessed by drawing from assembled experience of previous studies; even so, we must allow for changes over time since they were undertaken. But attrition rate is the most critical datum. Perhaps one might reasonably extrapolate from the experience of losses over time in the (meager?) assembled literature bearing on all relatively large long-term studies beginning at birth or soon after? Has anyone put together such material? Those data might yield at least a reasonable guesstimate of participation and loss rates within the earlier years of life (a decade at least in the NCCP, I think), especially if the data could be re-ordered in classes and proportions that resemble the expected distributions in a national sample.

In the end, this enterprise may come down to mustering the courage to sail into the unknown, with only the hope that material and other costs will be justified by the results. Of course, as others have also stressed, it is essential to frame the

whole undertaking in terms of what the hypotheses demand if they are to be adequately tested. But we can't brush aside the difficulties (nor the discomfiting thought of the billions at stake). Many a brave sailor, Captain Cook among them, perished on voyages into the unknown.

THIRD COMMENT 6-15 (RE JOHN LYNCH'S COMMENT OF 6-15)

Good! the discussion is advancing. Following up on it: 1) how about selecting a Center Sample to be representative of all regions across the United States, 2) then extending the study populations to separate representative Population Samples of women at risk of pregnancy (defined by age-group) in relatively well-defined areas around the selected Centers to provide supporting information on what is being missed.

FOURTH COMMENT 6-18 (RE PAUL SORLIE'S COMMENT OF 6-17)

Paul Sorlie's tables look good; they do make the case for centers as a base distinctly plausible, given community involvement as everyone seems to agree. I repeat the thought one might begin with center-based studies (perhaps stratified by size; there are I feel sure, more and better ideas about stratifying) randomly chosen across the country. These could be embellished by selecting supplementary random population samples in defined areas that more or less cover populations around those centers. We know that patients (or users) of medical centers are congregated mainly around the medical centers they use, however elite the institution. In both kinds of sample, if the information gathered at interview is to shed any light on physical and other aspects of environment, repeated home visits to examine housing conditions and physical environment, in both the short and the long run, will surely be necessary. Does this sound like the constructions in the late Rube Goldberg's cartoons?
Whatever decision is followed in the NCS, previous experience of longitudinal studies from birth tell us something about what retention and loss are to be expected etc. But do we have any basis in this country, beyond speculation, for estimating retention over time in a national random sample? I have not done a literature search; I can think of only one study that might fit the bill: if memory serves, Germaine Buck, before she moved from Buffalo to NICHD a few years ago, did do a study based on the follow-up of a (not very large) population-based sample. The only direct experience I can bring to mind is that of the British, beginning in 1946 with the National Children's Survey executed largely by James Douglas, and also it's successor initiated by the National Birthday Trust. If memory serves, the latter was begun in the late sixties and led by Neville Butler. In both these, national data on births were available and accessible and were statistically sampled and followed. Interesting although not, unfortunately, directly relevant for us beyond showing that, in the founding

English-speaking country a few decades back, national studies beginning at birth yielded important results.
In this country, the only study known to me that applied birth data in a partially national sample was a study of IQ in births to very young mothers, led and published by Zena Stein and Joy Dryfoos.They cobbled together the NCPP births and pegnancy data (center-based) and a National Health Survey (random sample), and were thus able to identify any skewing of the results in the NCPP cohort against the NHS data. So what is to be lost by a Center-based study in which selection bias can be measured against local population-based random samples? Yes, one will not have direct measures of exposures of interest before pregnancy or in very early pregnancy. Episodic events and the like will be missed but can be solicited, and no one is going to forget 9/11, or even traumas of much lesser degree. Stressful isolated or recurrent events are not difficult to elicit, and persisting stressful circumstances should as readily be tapped post-conception as before. The physical environment is generally a persisting element that can be repeatedly sampled to account for change over time; beginning from post-conception at registration, error in estimates should be minimal. Surely some of our many knowledgeable NCS participants will have more relevant US material than does this ramble into meager memory? As in my mind I worked through the questions and answers above, I came to the positive view in that the case for the hybrid approach I described was getting steadily stronger: we have sufficient existing data to make a respectable estimate of Center-based samples; there is really no doubt that the approach is a feasible one that has worked, growing better as epidemiologists advanced in technique; and we can provide reasonable guesses about attrition from the outset in a project adequately staffed and funded.
On the other hand, no pilot of a true population based sample can estimate attrition and other important issues over the intended life-course period proposed. If the hybrid design here suggested is rejected, and we simply (and unwisely) adopt the population-based approach holus-bolus, then given billions of $$$ at risk of disaster, I believe we are making a huge leap of faith. Unsupported by faith I have taken some large risks in my life-time, but the naked population-based sample makes me unaccustomedly nervous. It seems to me we could, and indeed should, be found culpable and rash if we do not beforehand proceed at least to discover what estimated losses would ensue in a preliminary substudy over a period of at least two years and preferably more.

FROM IRA TAGER

FIRST COMMENT 6-10 (RE FRANK SPEIZER'S COMMENTS OF 6-10)

I would like to amplify on Frank Speizer's comments. I am perplexed at the decision to use a probability sample not only for the complexity but related to the issues of exposures. In the latter case, a number of important environmental

exposures (air pollution, toxic chemicals in water, regional dietary habits) may not be sampled with sufficient numbers to provide adequately precise effect estimates. It seems to me that some center-based, target sampling has to be included. More importantly, to make these decision, one needs to have clear exposure outcome hypotheses. For the subjects for which I have expertise, I just do not see it. Based on the hypotheses with which I am familiar, I do not see a justification for a 21 year study that costs billions. To me, sampling issue remains secondary to the clarity of the science. If I were a congressman, I would not vote to fund the study as currently being conceived. Consequently, I cannot get myself into all of the subtleties of the sampling.

SECOND COMMENT 6-17 (RE PAUL SORLIE'S COMMENT OF 6-17)

I would like to add my "amen" to Paul's comments about the need for ongoing, local, community involvement to maximize follow-up and the need to consider this element in the selection of any sampling strategy.

FROM JANE TETA 6-11

I now have the advantage of responding after digesting the views of most of the SDWG. We find ourselves in the position of trying to develop a sampling scheme in the absence of clearly defined hypotheses. For more prevalent exposures, I prefer a national probablity sample and applaud the panel for raising the key advantages and disadvantages of this design and the center-based design and for recommending pilot studies. I think a strong nationwide and local communications plan and strong incentives, possible both financial and medical, would minimize recruitment and retention problems for a national probability sample. A pilot study would test my convictions. I also see no problem and definite advantages to the hybrid design.

The problem I see is with the national probability sample and hypotheses of environmental exposures of low prevalence or low level exposure - chemical or otherwise. For example, the majority of our knowledge in the area of chemical toxins comes from long duration, highly exposed workers or accidental overexposures in the general population. Dilution and imprecision would likely doom studies of low prevalence or low exposed members of the general population using a national probability sample. Such exposures are best investigated in targeted populations with common, high exposures. So the response to study design comes down to - which hypothesis?

BioMed Scientist JOBS

May 26, 2004

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A 500,000-person study?

Gene-environment interactions would be focus of NIH-led effort | By Maria W Anderson

The National Institutes of Health (NIH) is considering undertaking the largest population-based study ever done in the United States. NIH issued a request for information (RFI) from researchers earlier this month about the questions a large cohort study on the gene–environment interactions involved in common human diseases might ask, and how the study might be constructed.

A project of this kind is "the logical next step beyond the mapping of the human genome and doing case studies," said Terri Manolio, director of the National Heart, Lung, and Blood Institute's epidemiology and biometry program.

Such a project would try to survey a representative sample of the US population, explained Manolio, and may include as many as 500,000 participants from all geographic, racial, ethnic, and socioeconomic groups defined in the most recent US census. No funds have been appropriated for the project yet, and NIH officials are hesitant to speculate on how much it might cost.

Manolio said that NIH officials hope to find a way to incorporate data from previously conducted studies of individual diseases. "We want to include existing cohorts," she said, "but we have to decide, how feasible is it to add on to these disease studies?"

Alan Guttmacher, deputy director of the National Human Genome Research Institute (NHGRI), said that while there are questions about how the genotyping should be done—for example, whether it should all be done at once or if it should wait until the technology improves—identifying the environmental factors on which the study should focus, such as diet, lifestyle, and geographic area, might be the real challenge. "We don't have the expertise or the imagination to come up with all the hypotheses we want to answer with this data," he told *The Scientist*.

While the project could be likened to the UK BioBank and Iceland's deCODE Genetics, Guttmacher said, its objectives and approach would not be exactly the same. "The general idea is not dissimilar," he said, "but how we get there... would be different." For example, many of the minority ethnic groups that should be included in a US study are not present at all in the United Kingdom.

So far, the response from the research community has been generally positive, Manolio told *The Scientist*. "People are aware that there is room for something like this," Guttmacher said, adding that he has been "quite impressed" by the fact that scientists involved in similar research seem excited rather than threatened by the idea of this study.

"We know that a lot of genes contribute to [disease] risk, but aren't the only factor involved," said Terri Beaty, an epidemiologist at the Johns Hopkins University Bloomberg School of Public Health. A study of this kind could be "potentially very useful," she said, especially if we ever hope to attain the reality of personalized medicine.

NIH officials are unsure about how long the planning phase will last, how soon the project will get underway, and how soon it will start providing meaningful information. "Ideally, we will get useful data a few years into the study, but still be mining for information decades [later]," said Guttmacher. He said that a paper by NHGRI director Francis Collins explaining the benefits of such a study would be appearing in a major research journal later this week.

Although the official RFI closes this Friday (May 28), Guttmacher stressed that discussion of the project would be ongoing. Guttmacher said that the project, if initiated, would involve researchers from federal, academic, and private institutes, and that community involvement would also be a large component. NIH hopes to make as much as of the information freely available to the public as possible, which will require strict privacy guidelines.

NIH recognizes that a project of this magnitude would "cost a lot and take a long time," said Guttmacher, "but if you can't do it well, it's not worth doing... We're really trying to have the science design this study [and] drive the budget."

Beaty agreed: "It has a lot of potential, it needs to be done, and it needs to be done well," she told *The Scientist*.

Links for this article

"Request for information: design and implementation of a large-scale prospective cohort study of genetic and environmental influences on common diseases," National Institutes of Health press release, May 5, 2004. http://grants.nih.gov/grants/guide/notice-files/NOT-OD-04-041.h tml

T.M. Powledge, "Human genome project complete," *The Scientist*, April 15, 2003. http://www.biomedcentral.com/news/20030415/03

National Heart, Lung, and Blood Institute: Division of Epidemiology and Clinical Applications

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www.fidelitysystems.com

http://www.nhlbi.nih.gov/about/deca/

Alan Edward Guttmacher
http://www.genome.gov/10005495

P. Hagan, "UK Biobank reveals ethics framework," *The Scientist*, September 24, 2003.
http://www.biomedcentral.com/news/20030924/03/

Terri Beaty
http://faculty.jhsph.edu/?F=Terri&L=Beaty

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The case for a US prospective cohort study of genes and environment

Francis S. Collins

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Information from the Human Genome Project will be vital for defining the genetic and environmental factors that contribute to health and disease. Well-designed case–control studies of people with and without a particular disease are essential for this, but rigorous and unbiased conclusions about the causes of diseases and their population-wide impact will require a representative population to be monitored over time (a prospective cohort study). The time is right for the United States to consider such a project.

Identification of the genetic and environmental factors that contribute to health, disease and response to treatment is essential for the reduction of illness. This, of course, is the primary goal of biomedical research. Several auspicious recent developments suggest that progress in this area could be quite rapid. The sequence of the human genome[1,2] and increasing information about the genome's function have provided a robust foundation for the investigation of human health and disease. Likewise, results from the exploration of human genetic variation through the International HapMap Project[3] will soon furnish researchers with a powerful tool for identifying variants that contribute to common disease. This information will be especially useful when combined with reliable, cost-effective, high-throughput methods that can be used to genotype these variants in large population samples[4].

In parallel with the expansion of genomic tools and knowledge, methods for measuring non-genetic factors and environmental exposure have improved. These techniques promise to extend the range of epidemiological investigation[5]. There is growing recognition that a change in the environment, in combination with genetic disposition, has produced most recent epidemics of chronic disease, and may hold the key for reversing the course of some diseases[6]. For example, consider the interaction of presumed famine-protective genetic predispositions with a modern environment in which there is a ready availability of excess calories. This has probably contributed to the current obesity epidemic in the United States. Development of robust analytical methods for assessing disease-risk relationships and interactions is beginning to allow researchers to disentangle such complex effects on a population scale[7].

Together, these developments present an exciting opportunity to address unanswered questions related to the complex contributions of genes, the environment, and gene–gene and gene–environment interactions to

Rigorous quantitative assessment of genetic and environmental risk factors will be critical for the future of medicine.

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Box 1
Desirable characteristics of a gene–environment cohort study

To maximize the value of a prospective cohort study for determining gene, environment, gene–gene and gene–environment contributions to common disease, it should have many, if not all, of the following characteristics.

- A large number of participants, at least several hundred thousand, should be enrolled. This would ensure an adequate sample size for common disorders, particularly for gene–environment interactions.
- Minority groups should be intentionally over-sampled to permit meaningful inferences about these groups and for the study of health disparities.
- A broad range of ages should be represented to provide information on disorders from infancy to old age, with over-sampling of age groups as needed.
- A broad range of genetic backgrounds and environmental exposures should be included to provide enough variability to detect and compare associations and interactions.
- Family-based recruitment, including multiple generations, should be used for at least part of the cohort to increase the power of genetic analyses.
- A broad array of clinical and laboratory information, not limited to any single disease, should be collected at the beginning and at regular intervals thereafter.
- Sophisticated dietary, lifestyle and environmental exposure assessments should be carried out, using both questionnaires and biological measures.
- Biological specimens, including DNA, plasma and cells, should be collected and stored.
- A highly sophisticated data-management system should be included.
- Access to study data and biological materials should be free and open to allow research into many diseases by scientists in many sectors.
- Investigations during the study should not be limited to hypotheses conceived at its inception.
- Comprehensive community engagement should be a major feature in the design and implementation of the study.
- A state-of-the-art consent process should be adopted to allow multiple uses of the data and regular feedback to participants about progress.

health. Understanding these factors and their interactions could lead to major improvement in diagnostics, preventive medicine and therapeutics.

Case–control studies and beyond

A widely used and highly successful approach to identifying factors that contribute to specific illnesses is the case–control study. For this, carefully chosen people with and without a disease are analysed for differences in the distributions of genetic variation and/or environmental exposures or other non-genetic factors[8]. Valuable insights, perhaps unobtainable in any other way, have been derived from such studies, particularly for rare disorders. False-positive genetic associations related to differences among various population groups have been a problem in the past, but the availability of high-throughput, low-cost genotyping can reduce this risk by pre-matching genetic markers with a panel of random ones or otherwise adjusting for background genetic differences[9].

Case–control studies have certain weaknesses, including the tendency for clinically diagnosed cases to represent the more severe end of the disease spectrum[10] and the difficulty of selecting an unbiased control group. In addition, many case–control studies are plagued by the problem of recall bias: memories of individuals diagnosed with disease are often coloured by their subsequent experience of illness[11]. For example, a recent case–control study of coronary heart disease showed that people with heart disease were more likely to report a family history of the disease that could not be verified than were controls[12]. Although this problem may not affect genotype-specific risk-ratio calculation[13], it is still a significant problem for the overall assessment of disease risks.

So, although the case–control strategy can be a powerful means for identifying potential risk factors, its inherent biases make the quantification and population-wide generalization of risk difficult. Replication of associations and estimation of their magnitude, consistency and temporality (all key criteria for epidemiological evidence of causal relationships[14]) are best obtained through prospective, population-based cohort studies[8].

To appreciate the contrasting but potentially complementary nature of case–control and prospective cohort studies, consider the example of diabetes. A case–control study of 5,000 cases and 5,000 controls could be mounted over a year or two, and could be used to identify susceptibility genes and environmental correlates of risk. But selection biases for the phenotype (for instance, a previously known diagnosis of diabetes) would prevent quantitative generalization of the results. Furthermore, such a study would probably be subject to recall biases among the cases about their family history, diet and other environmental factors, and there would be no specimens available from the people being studied before diagnosis to search for predictive biomarkers. These shortcomings could be addressed by a longitudinal study of 200,000 people, but it would probably take several years for 5,000 of them to develop diabetes. In the long run, however, the need for this kind of information for 40 common diseases would require the collection of data on 200,000 people anyway, and the prospective cohort study would also allow links to other conditions (such as hypertension and obesity) to be detected.

Identifying logistical hurdles

Along with the many advantages of prospective studies is a unique set of challenges, most of which centre on logistics. Such studies generally require large sample sizes, detailed characterization at the beginning of the study, and prolonged follow-up for the occurrence of most common chronic diseases[8] (see Box 1).

Large-scale cohort studies are under discussion or already underway in the United Kingdom (the UK BioBank), Iceland (deCODE), Estonia, Germany, Canada and Japan. Although such projects are likely to be useful for research everywhere, the United States should seriously consider undertaking a national investigation of its own. Inadequate representation of important US minority groups who bear disproportionate burdens of disease (particularly African-Americans, Latinos and Native Americans), the probable presence of different environmental risk factors, and the potential for limited access to data and biological materials make it unlikely that the current cohort projects will be adequate for the needs of the United States.

In the United States, a gene–environment cohort study could be assembled by building, at least in part, on already existing large studies such as the Women's Health Initiative, the Framingham Study, the Harvard studies of health professionals, and some of the many large cancer cohorts. The obvious advantages are that many years of follow-up have already taken place in these cohorts and, for many of them, DNA has already been collected. But serious consideration must be given to whether the disease-specific focus of many of these studies has limited the phenotyping and exposure measures, whether the minority representation is adequate, whether the consent obtained is sufficient for broad access to data and biological materials, and whether the study design is appropriate for the

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ambitious goals of a national gene–environment study. If those limitations turn out to be significant, an entirely new cohort project may need to be contemplated.

Evaluating the merits

Although the challenges in undertaking such a prospective population study in the United States will be considerable, a serious evaluation of its merits is now in order. This debate should engage a wide variety of experts in epidemiology, genetics, environmental science, ethics, public health, economics and public policy. An initial meeting at the National Institutes of Health in December 2003 led to agreement that such an effort should be explored further. If the conclusion is that this resource is needed, then we must collectively seek ways to organize and implement it quickly and efficiently — or face the real possibility that a decade from now the promise of genetic and environmental research for reducing disease burden on a population basis will remain out of reach. □

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Mr. Rolter is a management and organizational development consultant with extensive experience assisting with organizational design and implementation of strategic and operational changes to improve business systems, performance and effectiveness. Over the past several years, he has assisted numerous private, public sector, and not-for-profit organizations in North America and Europe to:

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In the private sector Mr. Rolter has assisted a variety of organizations from Fortune 500 corporations to a start-up firm, in a various industries including; Oil & Gas, Telecommunications, Banking and Real Estate, Publishing, Aerospace, Manufacturing, Professional Services, Personal Services, and non-profit Associations. In the public arena, Mr. Rolter has consulted to regulatory, oversight and service delivery organizations design and implement change programs for both single and cross agency initiatives at the Federal and local levels.

Mr. Rolter focuses on helping client organizations improve performance by combining management consulting concepts and organizational development approaches to mobilize the client organization, achieve performance changes, and ultimately modify the organization's culture. Mr. Rolter utilizes a comprehensive approach tapping into management, employee, and customer perspectives to deliver practical programs for improving organizational performance and facilitating client teams to accomplish them. He has guided and facilitated client organizations through the design and implementation of major strategic and operational change programs including performance measurement, business process re-engineering, application of new technology, restructuring, productivity improvement, strategic planning, activity based costing, process management, and other enterprise-wide business changes.

Mr. Rolter has over 20 years of professional experience that began as an engineer and technologist with McDonnell Douglas Astronautics. Subsequently, Mr. Rolter performed technology sales and services for Hewlett-Packard, and management consulting services for Booz-Allen & Hamilton, American Management Systems, and Arthur Andersen & Co.

Mr. Rolter has delivered numerous presentations and real-time experiential training courses focused on implementing changes in organizations, reengineering, benchmarking, team development, performance metrics, 6 Sigma and quality management, leadership, presentations, program development, and other business concepts and tools. He has also acted as a coach to advise and develop clients and other consultants in creating and applying approaches, tools, and techniques for organizational analysis, implementation, and managing change efforts in organizations.

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- Process Management and Improvement
- Organizational Analysis & Assessment
- Organization Development & Change Management
- Quality Management
- Strategic Planning
- Business Process Reengineering
- Project Implementation & Management
- Benchmarking
- Team Facilitation

Mr. Rolter resides in Arlington, Virginia and holds a Masters of Business Administration from the Anderson School of Business at UCLA, a Bachelor of Science in Mechanical Engineering from Cornell University, and is a graduate of the Organizational Development and Change Management Program at Georgetown University in Washington, DC. He is affiliated with the Organizational Development Network, the International Association of Facilitators, and the Strategic Leadership Forum.

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National Children's Study Meetings
As of 06/16/04

Workshop: Community Engagement
December 3, 2002
Natcher Center, NIH, Bethesda, MD
Organized by: Community Outreach and Communications Working Group
Contact: Diane Dennis-Flagler

Workshop: International Consultation on Longitudinal Cohort Studies
December 16, 2002
Baltimore Marriott Waterfront, Baltimore, MD
Organized by: Special Committee
Contact: Adolfo Correa, Danuta Krotoski

W16 Workshop: Fetal and Neonatal Growth and Development Workshop
December 15–16, 2002
Baltimore Marriott Waterfront, Baltimore, MD
Organized by: Pregnancy and the Infant Working Group
Contact: Adolfo Correa

Workshop: Medicines Exposures: Collection, Coding, and Classification
December 16, 2002
Baltimore Marriott Waterfront, Baltimore, MD
Organized by the Medicine and Pharmaceuticals Working Group
Contact: Diane Kennedy

Federal Consortium Meeting
December 17, 2002
Baltimore Marriott Waterfront, Baltimore, MD

NCSAC Meeting
December 17–18, 2002
Baltimore Marriott Waterfront, Baltimore, MD

Study Assembly, Working Group Meetings, and Inter-Working Group Meetings
December 17–18, 2002
Baltimore Marriott Waterfront, Baltimore, MD

NCSAC Meeting
March 6–7, 2003
NICHD, Rockville, MD

W2 Workshop: Innovative Technologies for Remote Collection of Data for the National Children's Study
May 12–13, 2003
Boston, MA
Organized by: U.S. Environmental Protection Agency

ICC Retreat
May 19–22, 2003
Founders Inn Conference Center, Virginia Beach, VA

W9 Workshop: Ethical Issues in Longitudinal Pediatric Studies: "Looking Back, Thinking Forward"
June 4, 2003
Holiday Inn Select, Bethesda, MD
Organized by: Ethics Working Group
Contact: Ben Wilfond, Jeff Botkin

NCSAC Meeting
June 5–6, 2003
Holiday Inn Select, Bethesda, MD

W24 Workshop: Assessing the Incidence and Outcomes of Mild Traumatic Brain Injury in the National Children's Study
September 11–12, 2003
Holiday Inn Select, Bethesda, MD
Organized by: Injury Working Group
Contact: Gitanjali Saluja, Ruth Brenner

NCSAC Meeting
September 15–16, 2003
(Working Dinner September 14 at 6:30 p.m.: NCSAC members only)
Holiday Inn Select, Bethesda, MD

Working Group Meeting: Exposure to Chemical Agents
October 28–29, 2003
American Chemistry Council, Rosslyn, VA
Organized by: Exposure to Chemical Agents Working Group
Contact: Haluk Ozkaynak

W26 Workshop: Placental Measurements
November 3–4, 2003
Holiday Inn Select, Bethesda, MD
Organized by: Early Origins of Adult Health Working Group
Contact: Ken Schoendorf, Catherine Spong

W10 Workshop: Psychosocial Stress and Pregnancy and Infancy
November 12–13, 2003
Holiday Inn Select, Bethesda, MD
Organized by: Pregnancy and the Infant Working Group
Contact: Marian Willinger, Mark Klebanoff

W33 Workshop: Measuring Physical Activity in the National Children's Study
November 17–18, 2003
Crystal City Marriott, Arlington, VA
Organized by: Special Committee of Interagency Coordinating Committee and Working Group Members
Contact: Amy Branum, Mary Hediger

Workshop: Pilot Study Review
November 21, 2003
EPA, Research Triangle Park, NC
Organized by: Interagency Coordinating Committee
Contact: Carole Kimmel

NCSAC Meeting
December 15–16, 2003
Sheraton Atlanta, Atlanta, GA

Working Group Meeting: Birth Defects
December 15–16, 2003
Sheraton Atlanta, Atlanta, GA
Organized by: Birth Defects Working Group
Contact: Cheryl Hobbs
MEETING CANCELLED

Working Group Meeting: Community Outreach and Communications
December 16, 2003
Sheraton Atlanta, Atlanta, GA
Organized by: Community Outreach and Communications Working Group
Contact: Diane Dennis-Flager

Working Group Meeting: Health Disparities and Environmental Justice
December 16, 2003
Sheraton Atlanta, Atlanta, GA
Organized by: Health Disparities and Environmental Justice Working Group
Contact: Kristine Suozzi

Working Group Meeting: Ethics Working Group
December 16, 2003
Sheraton Atlanta, Atlanta, GA
Organized by: Ethics Working Group
Contact: Ben Wilfond, Jeff Botkin

W28 Workshop: Use of Herbal Products in Pregnancy, Breastfeeding, and Childhood
December 16, 2003
Sheraton Atlanta, Atlanta, GA
Organized by: Medicine and Pharmaceuticals Working Group
Contact: Diane Kennedy

Study Assembly Meeting
December 17, 2003
Sheraton Atlanta, Atlanta, GA

W30 Workshop: Media Effects on Child Health and Development
Date and Location: January 22–23, 2004
Renaissance Austin, Austin, TX
Organized by Social Environment Working Group
Contact: Christine Bachrach

Working Group Meeting: Social Environment Working Group
Date and Location: February 10–11, 2004
Holiday Inn Select, Bethesda, MD
Organized by Social Environment Working Group
Contact: Christine Bachrach

Working Group Meeting: Health Services Working Group
Date and Location: February 26–27, 2004
Holiday Inn Select, Bethesda, MD
Organized by: Health Services Working Group
Contact: Denise Dougherty

W31 Workshop: Addressing Rural Children in the National Children's Study
Date and Location: March 2, 2004
Holiday Inn Select, Bethesda, MD
Organized by: Social Environment Working Group
Contact: Christine Bachrach

Working Group Meeting: Exposures to Chemical Agents Working Group
Date and Location: March 3, 2004
Holiday Inn Select, Bethesda, MD
Organized by: Exposures to Chemical Agents Working Group
Contact: Haluk Ozkaynak

Working Group Meeting: Injury Working Group
Date and Location: March 3, 2004
Holiday Inn Select, Bethesda, MD
Organized by: Injury Working Group
Contact: John Lutzker

NCSAC Meeting
March 4–5, 2004
Holiday Inn Select, Bethesda, MD

W29 Workshop: Sampling Design
Date and Location: March 21–22, 2004
Sheraton Crystal City, Arlington, VA
Organized by: Special Committee of Interagency Coordination Committee, NCSAC, and Working Group members
Contact: Jim Quackenboss

ICC Retreat
April 15–16, 2004
Doubletree Hotel and Executive Meeting Center, Rockville, MD

Working Group Meeting: Birth Defects Working Group
Date and Location: April 15, 2004
Embassy Suites, College Park, GA
Organized by: Birth Defects Working Group
Contact: Charlotte Hobbs

Healthy Development Ad Hoc Working Group Meeting
Date and Location: May 4, 2004
Palace Hotel, San Francisco, CA
Organized by: Special Committee of the NCSAC
Contact: Neal Halfon/Paul Wise

W12 Workshop: Expanding Methodologies for Capturing Day-Specific Probabilities of Conception
Date and Location: May 17-18, 2004
Doubletree Hotel Rockville, Rockville, MD
Organized by: Fertility and Early Pregnancy Working Group
Contact: Warren Galke/Joseph Stanford

W34 Workshop: Cancer and the National Children's Study: Opportunities and Challenges
Date and Location: May 20, 2004
Holiday Inn Select, Bethesda, MD
Organized by: Interagency Coordinating Committee and Program Office
Contact: Peter Scheidt and Rebecca Brown

W36 Workshop: Measurement of Maternal and Fetal Infection and Inflammation Workshop
Date and Location: May 20–21, 2004
Embassy Suites Hotel Baltimore at BWI
Linthicum, MD
Organized by: Interagency Coordinating Committee
Contact: Ken Schoendorf

W22/23 Workshop: Methods for the Assessment of Asthma-Related Health Outcomes
Date and Location: May 27–28, 2004
Rosen Centre Hotel, Orlando, FL
Organized by: Asthma Working Group
Contact: Pauline Mendola

W37 Workshop: Gene Environment Interaction and the Regulation of Behavior
Date and Location: June 2–3, 2004
Holiday Inn Select, Bethesda, MD
Organized by: Development and Behavior and Social Environment Working Groups
Contact: Sarah Knox

Sampling Design Subcommittee Meeting
Date and Location: June 4, 2004
NICHD, Bethesda, MD
Organized by: Special Committee of Interagency Coordinating Committee, National Children's Study Program Office, and the NCSAC
Contact: Jim Quackenboss and Jan Leahey

Workshop: Measuring Racial/Ethic Disparities and Racism from a Developmental Perspective Workshop
Date and location: June 21–22, 2004
Doubletree Hotel Rockville, Rockville, MD
Organized by: Health Disparities and Environmental Justice Working Group
Contact: Sarah Knox

NCSAC Meeting
June 28–29, 2004
Holiday Inn Select Old Town, Alexandria, VA

W53 Workshop: Measures of Neurobehavioral Development and Environmental Exposures
Date and location TBD
Organized by: Social Environment Working Group
Contact: Carole Kimmel/Tracey Thomas

W38 Workshop: Assessing Dietary Intakes and Patterns in Women and Young Children: Methodological Issues with Implications for the Design of the National Children's Study
Date and Location: TBD (Suggested: Fall 2004)
Organized by: Early Origins of Adult Health Working Group
Contact: Adolfo Correa

Workshop: Body Composition Measurement for the National Children's Study
Date and location: TBD (Suggested: October 7–8, 2004, DC area)
Organized by: Nutrition, Growth, and Pubertal Development Working Group
Contact: Mary Hediger

NCSAC Meeting
September 27–28, 2004
Location TBD

NCSAC Meeting
December 9–10, 2004
Location TBD

THE NATIONAL CHILDREN'S STUDY ADVISORY COMMITTEE
2004 MEETING SCHEDULE

September 27–28, 2004	Advisory Committee Only; Location TBD
December 9–10, 2004	Advisory Committee Only; Location TBD

National Children's Study
Community Outreach and Communications Working Group
Guidance Document
June 18, 2004

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Executive Summary

The National Children's Study's (Study) main objective is to examine the environmental influences on children's health and development. The Study will explore a broad range of environmental factors to discern helpful or harmful influences on the health and well-being of children. The aim is to better understand the role of these environmental factors and therefore provide a more effective basis for health promotion and health care practices. The Community Outreach and Communications Working Group (COC), as well as a growing number of scientists and other health care professionals, agree on the collaborative model for community research. Studies have shown that one of the most effective ways to produce relevant and efficacious research results is to involve the research participants throughout the full life cycle of the project. This approach:

- Includes the community in the design and planning of the Study
- Establishes a vehicle for interaction between the researchers and the community
- Retains participation and interests based on solicited input from the community
- Provides periodic updates on the status/progress of the research
- Leaves the community in a better position than when initially encountered (for example, capacity building).

The COC recognizes the challenges of undertaking meaningful community participation in a study of this magnitude. This national longitudinal Study will include different geographical areas and persons residing in communities that vary among ethnic and cultural dimensions. Ironically, such a challenge also gives the Study design its richness. The COC has expressed concern that currently there appears to be no structural provisions for the inclusion of community comment or meaningful involvement in the shaping of the Study other than using community members as Study subjects. This working group is eager to assist in establishing and/or facilitating such a vehicle.

Some costs/investments and benefits to using the participatory research model are as follows:

Cost/Investment	Benefit
▪ Gain community "buy in"	▪ Mutual trust
▪ Help retention efforts	▪ Reliable data
▪ Build community capacity	▪ Community trust and respect; making a tangible contribution to the community would facilitate the possibility of future research efforts
▪ Disseminate appropriate Study updates	▪ Maintain interest, retain participants, and assist in continued recruitment

A few of the greatest benefits of having community participation in the planning stages and throughout the Study are:

- Collaboration between researchers and the community in each stage of the Study will help to better identify and define health problems, environmental exposures (social and physical)

that may be important to child health and development, and other associated issues, which will ultimately produce better data.

- Community participation provides an opportunity for reciprocal education between the community and researchers to help both gain a better understanding of the whole picture.
- Partnership with the community facilitates implementation of the Study by assisting to both identify and find acceptable solutions to ethical issues, and by assisting in the design and dissemination of appropriate information about and results from the Study.
- Community input and planning throughout the Study builds community support and capacity, and fosters a level of community trust and respect that would facilitate better recruitment and retention, strengthen commitment to the project over the life of the Study, and increase the possibility of participation in future research efforts.

This document is by no means complete. The COC considers this a "living document," and as more of the Study design is made clearer, this document will respond with strategies appropriate for implementing and involving the community in a meaningful way. Once the Study is underway, the value of applying a well-designed, comprehensive community outreach strategy cannot be understated. There is a critical need to consider culturally specific approaches and concepts in order to respond to the needs and values contained in representative American communities.

Introduction

The Study's main objective is to study the environmental influences on children's health and development. The Study hopes to explore a broad range of environmental factors, both helpful and harmful, that influences the health and well-being of children. The aim is to better understand the role of these environmental factors on the health of children and therefore provide a more solid basis for effective health promotion and health care practices. The COC, as well as a growing number of scientists and other health care professionals, believe that one of the most efficient ways to produce relevant and efficacious research results is to involve the research participants not only as subjects of the study but also in the actual design and planning of the study.

The Study recognizes the importance of meaningful community feedback and participation into the design and execution of this Study. However, the challenge is how to incorporate that feedback into the Study design and other processes of the Study. In an effort to illustrate some of the strategies and methods that can be used to accomplish this task, and to avoid "reinventing the wheel," the COC felt that borrowing ideas and concepts from community-based research efforts would be a good place to start. Because of the magnitude and time this Study will take to complete, the ability to retain participation in the Study over the 20-year period will be predicated on the skill in promoting trust and in building capacity within the communities through empowerment.

Document Purpose and Description

This document outlines and gives general guidance and support on how the Study should and can incorporate meaningful community participation into the design and implementation of the Study. Given the still unfolding nature of the Study design, this document is still a work in progress. As a consequence, some sections will not be completed until decisions about the design of the Study are complete. Therefore, the COC has divided the document into three major sections:

- Background, benefits of meaningful community participation, and community engagement
- Presite selection recommendations
- Postsite selection recommended strategies.

The information and evidence presented in this document should encourage the National Children's Study Advisory Committee (NCSAC) to accept the COC's recommendations and support the working group in persuading the planning committee to include local community participation in the planning and implementation of the Study. Once the targeted communities are identified, the COC can work on tailoring communication strategies specific to the targeted communities and support the other working groups, such as the Study Design Working Group, in the best ways to incorporate community input into their processes.

The information contained in this document is compiled from three main sources:

- Review of the literature
- An expert panel roundtable discussion held in December 2002
- The experience of the COC members.

Background, Benefits of Meaningful Community Participation, and Community Engagement

Background. Over the past 30 years, participatory research has been underway throughout the world. The scientific research community has begun to realize that researchers need to be more sensitive to the need of the general public to be involved in the development of health promotion and health care practices. For this very reason, participatory or some type of community-based research, which includes meaningful input from the targeted groups in some of the decision-making processes, is crucial to the success of the Study. Participatory research is becoming increasingly important in the health care field because communities want to take greater ownership and control over decisions affecting their health. Participatory research has been growing in prominence because of the communities' need for control and empowerment.

Unfortunately, health research has seen some dark moments over the years. Science researchers have involved the human population in various experiments and studies of one type or another. Sadly, the involvement has not always been fully consensual or the dangers or risks involved have not been fully disclosed. As a result of these types of research studies, communities are wary of research scientists and suspicious of their research agendas.[1] Therefore, the Study has quite a bit of history to overcome as it tries to engage, recruit, and retain participants.

On the other side, researchers have some valid reasons for not avidly seeking community input during the formative planning and designing phases of research studies. Developing rapport and building trust in communities is at times challenging and very time consuming. In addition, research scientists may feel that inclusion of the targeted communities in the planning and designing phase of the Study may compromise the integrity of the science. These reasons are understandable, but there are ways to address those issues and still allow communities to participate in the process from the start. Exploring ways to overcome those issues and providing a balance between the rigor of scientific research and the communities' empowerment needs is one of the purposes for this document.

Benefits of Meaningful Community Participation. Despite the recognition of the benefits of a participatory approach for the Study, the COC recognizes the challenges of undertaking meaningful community participation in a study that will be undertaken across many different geographical areas and with persons residing in communities that vary along ethnic and cultural dimensions. Another challenge to community engagement at this point in the Study is the still unknown nature of the Study hypotheses and, consequently, the Study design. The COC has expressed concerns that currently there seems to be no structural provisions for the inclusion of community comment or involvement in the shaping of the Study other than using community

members as Study subjects. The Study is currently reviewing a number of possible Study hypotheses.

Although the exact Study hypotheses have not yet been identified, we do know that some will involve the collection of body fluids and tissue samples. Although it may be possible to elicit consent on a one-time or limited basis for such collections, to retain cooperation and the continuation of such collections over an 18- to 20-year period necessitates a real commitment and consensus of the Study participants. The best way to ensure that commitment is by allowing the studied groups to have a part in the planning of the research so that participants will have a better understanding and sense of ownership and will stay with the process over the long haul.

Participation of community representatives will be instrumental in helping to decide appropriate recruitment strategies (including selection of incentives) and ensuring that informed consent takes place. Community representatives can be extremely valuable in suggesting strategies for publicizing the Study in general and advising researchers on the drafting of recruitment materials to explain the Study and the benefits and commitments the Study may entail.[2] The Study may be planning to supply all sites with standardized recruitment information and brochures but the COC hopes that there will still be latitude for individual sites to work with their community advisory boards (CABs) to develop materials that may be more relevant to the context of that site.

Community representatives can also be useful in reviewing informed consent procedures and documents and suggesting modifications to these documents to ensure that participants are truly informed and aware of their rights and responsibilities in terms of the Study. Certainly, their suggestions will have to be considered in light of legal and other ethical considerations but they can serve as a valuable resource to the sites and the overall Study institutional review board.

Although there has been discussion about standardizing incentives for all Study participants, the COC suggests that a “one size fits all” approach should not be used; instead, community representatives should be involved in deciding appropriate incentives for the participants from their communities.

In recent years, there has been increased call in the health and social sciences for research that involves community representatives in all aspects of the research. This type of research has been referred to by different names including “community-involved research,” “community-centered research,” “researcher–constituent collaboration,” and “community-based participatory research.”[3]

Of particular importance to the Study, are the suggestions that engagement and participation of community representatives in all aspects of the research can enhance the quality of the research. Engagement will not only increase the likelihood that community members will agree to be participants in the research, but the actual research questions themselves (and subsequent data collection activities) will more likely reflect the actual social and physical environmental

influences to which the children are exposed. In a community-based participatory research (CBPR) model, community members will also be involved at the beginning of the research in helping to define what should be "studied" in the research.[3] Current examples of children-centered research that have used a CBPR model to engage community members in all aspects of the research are the EPA and the National Institute of Environmental Health Sciences-funded Centers for Excellence in Children's Environmental Health.[4]

Achieving the goals of community engagement depends on the active involvement of a range of stakeholders working together as representatives of communities. Their involvement needs to be authentic and should occur as early as possible in the process.

Community Engagement. Community engagement in research can be thought of along a continuum centered on the extent to which there is the participation and influence of nonacademic researchers in all phases of the research. On one end of the continuum are research projects that emphasize community as a place or setting and involve community members primarily as research "subjects." On the other end of the continuum are research projects that emphasize community as a social and cultural entity and include the active engagement and influence of community members in all aspects of the research project.

Some researchers, citing social science literature, suggest community is characterized by a sense of identification and emotional connection to other members, common symbol systems, shared values and norms, mutual (although not necessarily equal) influence, common interests, and commitment to meeting shared needs.[3] Others define community as a group of people with diverse characteristics who are linked by social ties, shared common perspectives, and engaged in joint action in geographical locations or settings.[5] This definition emerged from focus group interviews at four sites across the country. As noted by the authors, this definition parallels similar social science definitions of community that, according to the authors, confirms the viability of a common definition for participatory public health. Based on the results of this research and a review of the literature, the COC suggests the Study consider a definition of community that includes attention to social ties, common perspectives, and a shared sense of identification and not just a common geographical location.

Some factors for the Study planners to consider are:

- Become knowledgeable about the community in terms of its economic conditions, political structures, norms and values, demographic trends, history, and experience with engagement efforts. In our expert panel workshop, several participants suggested creating a community profile together with community members to determine what they perceive as ailing them in order to design mutually beneficial site-specific questions.
- Establish relationships, build trust, and collaborate with formal and informal leadership to seek commitment from community organizations and leaders to create processes for community participation in the research.
- Be cognizant of the language used. Members of the expert panel stressed the need to clearly define all terminology used to create a standardized understanding of what is meant among

researchers and community members. The expert panel also suggested that researchers avoid language that marginalizes or make communities "other," (for example, "we, us, ours" versus "you, they, or your") or can have pejorative connotations ("participant" versus "subject").

- Recognize and respect community diversity. Awareness of the various cultures of a community and other factors of diversity must be included in designing and implementing not only the research but also the strategy for community engagement. This requires researchers to exhibit cultural sensitivity and competence in their interactions with community members. While the inclusion of persons of color on the research team may help to increase the cultural awareness of the research team, the expert panel warned the Study to be wary of the myth that "minority scientists" can gain community buy-in by virtue of their race/ethnicity/nationality alone.
- Make a long-term commitment to work with the community in feeding back the data and possibly designing interventions based on Study data.

Presite Selection Recommendations

The initial contact made between the communities and the Study is vitally important to the Study's successful entrée into the community. The COC and experts from the workshop suggest the Study aggressively pursues getting the word out about the Study so that potential participants will become familiar with the Study, its purposes, and how it relates to them. The COC recommends the following three-step approach to this initial contact with communities:

- Market the Study to the public.
- Identify national organizations that sponsor or operate social and health-related programs in local communities across the country, as well as local organizations of influence in the community.
- Engage representatives from local communities in an official capacity.

Step 1: Marketing the Study. The COC suggests a social marketing strategy to accomplish the goal of getting the message out to communities. Social marketing could also be used for the life of the Study to continue promoting the Study and sustaining awareness of its ultimate goals. Social marketing as a discipline was born in the 1970s when it was realized that the same marketing principles that were being used to sell products to consumers could be used to "sell" ideas, attitudes, and behaviors. Research is crucial to determine the most effective and efficient vehicles to reach the targeted audiences and get the message across. This is why it is crucial to involve the types of people to be targeted in the planning process and development of strategy. Examples of marketing efforts include the following:

- **Identity branding.** Branding will help to establish a distinct identity for the Study. Development of logos and other designs creates a "look and feel" for the Study project, which will make it easily recognizable to the public. Some of this has already started with the development of the Study "children's" logo.
- **Use of mass media in outreach efforts.** Mass media outlets such as radio, newspapers, and television are obviously good sources for dissemination of information about the Study. Yet, relying on public service announcements as the source for dissemination of the information

to these outlets may result in the message being buried in the newspaper or being aired at times people may not be watching or listening. Contacting the media directly to ask them to do a story on the Study and including in that story how community members can become involved is one strategy that might work with the media outlets. In identifying media outlets to work with, one should remember the smaller local newspapers that might focus on a specialized market. For example, many metropolitan cities have newspapers and/or radios that focus on African Americans, Latinos, or other ethnic groups.

- **Organizational newsletters and mailings.** Writing articles for organizational newsletters and mailings is another way to disseminate information about the Study to the public. Many community-based organizations (CBOs) and agencies have newsletters that are published quarterly or even monthly. These agencies are often happy to include information if provided with an already drafted article by their publication deadline. Once established, Study sites may want to start their own newsletter as a way to disseminate information about the Study to participants and the public.

Step 2: Partnerships and Coalition Building. Due to the scope and magnitude of the Study, it will need to collaborate with other organizations in the community. It will be important to align with organizations that demonstrate an understanding of the community, some of which maybe national in scope while others maybe local. Recognized organizations that run community health and social programs, such as the March of Dimes, United Way, and National Urban League may provide valuable input into strategic marketing of the study. In addition to these groups, local grassroots groups such as church organizations, local civic clubs, sororal and fraternal organizations, etc. should also be involved to ensure suitability of the marketing efforts. Collaborating with these community organizations to develop community profiles enhances understanding of the communities and facilitates development of communication strategies. In addition, liaisons with faith-based organizations, schools, and other organizations could also be instrumental in the development of the community profile, which would give one a better sense of the community dynamics. This gives Study planners the benefit of knowledge, experience, insight, and rapport with the community. The profiles can then be used later to help locate and identify individual community members to represent the community on a CAB.

A common process often used to engage communities in research is to identify and engage community leaders. This usually takes the form of some type of community advisory committee. This committee normally does not include participants in the Study, but instead includes representatives of the communities in which the participants reside. Advisory committees are asked to represent the interests of both the participants and the broader communities.

Community advisory committees can vary greatly in their participation and influence in the research process. Some researchers distinguish advisory committees from steering committees suggesting the latter have more direct control over the decisions of the research process.[3] For example, the National Center for Early Development and Learning increased constituent collaboration with researchers because it wanted to “go beyond focus groups and advisory boards” and have “constituents collaborate actively with researchers to identify the kinds of

research information needed by parent, teacher, and other consumers and help determine the best ways to disseminate information."[6]

The expert panel suggested that these advisory committees could provide valuable insight into relevant issues that the researchers may not have considered and advocated for a "true partnership" approach with communities in which their advice is actively incorporated into the research design and implementation. The panel warned that the Study must be upfront about the amount of community engagement it will accept because there is a broad continuum between the desire to engage the community for better recruitment and the desire to engage the community in all aspects of the research. Misleading community representatives about the amount of their influence and participation may affect the trust and eventually the Study itself.

Some researchers stress the need to make sure members of the advisory committee are connected to the people in their community and represent some type of constituency from that community. These researchers cite many examples of how advisory boards can effectively represent communities if the right representatives are chosen:[5]

> Based on our experiences and our data, an important element for success may be ensuring that CAB representatives are actively connected to diverse people in their local communities and empowered to function in ways that are meaningful to their community base. Other research supports this view. Conway and colleagues [7] compared perceptions of health priorities among local District Health Council members and among a random sample of household residents in Chicago and Cook County, Illinois. The results showed substantial agreement in priorities, indicating that advisory boards can effectively represent community perspectives regarding health priorities. Jewkes and Murcott [8] presented results of a qualitative assessment of the uses, meanings, and interpretations of community participation in the context of the World Health Organization's Healthy Cities Project as implemented in the United Kingdom. In interviews with 50 participants drawn from health, local government, and voluntary sectors, they found that "being known" was the most fundamental requirement of an effective representative. Data from a case study by Bond and Keys [9] p37, support the feasibility of empowering multiple community groups simultaneously through a single advisory board "when the board culture promoted inclusionary group processes and the activation of member resources."

Often CABs consist of representatives from CBOs or other local agencies. A downside of this approach is that it may omit representation by participants who are not constituents of the CBOs and the agencies represented. In addition, as members of the expert panel pointed out, communities are not monolithic structures; and even within similar ethnic or cultural groups, there exists a great deal of within-group variability. Thus, Study researchers need to remember that data collected based on community needs does not necessarily reflect an individual's needs.

The COC is recommending a two-tier approach to the CAB process. We propose that the first tier include representatives from national organizations. The representatives must have enough influence and be strategically placed within their respective organizations so that they can

command the attention of the decision makers and garner support and/or resources that might be needed once the Study is underway. The second tier of the CAB would consist of representatives from the selected Study areas.

Step 3: Preliminary Engagement/Outreach. After establishing local level collaborative partnerships, the Study should seek opportunities to meet with representative community members to communicate the aims of the Study and to solicit feedback. A variety of strategies can be used to engage communities in the Study. Some of initial strategies will be executed centrally by the Study, such as the introductory publicity campaign that includes the distribution of brochures and press releases to policymakers and other potential stakeholders.

In the early stages of recruitment for the Study, public meetings and presentations to explain the Study should be considered. The feasibility of public meetings may depend on the type of sampling design that is ultimately chosen. For example, if a simple random sample is chosen in which participants come from a large geographical area such as a state, then one or two public meetings may not be sufficient. However, in that case, public presentations in which the Study is explained to representatives of the media may be a useful strategy. If public meetings are held, conveniently scheduled meetings should be announced well in advance to encourage broad participation.

The sampling design for the Study may not sample based on communities as an entity but instead may undertake a simple random sample or some type of cluster sample within a larger geographical area (such as a state). Obviously, it is a much more difficult task to engage communities in a research process when the actual number of residents involved in any one community may be small and when the number of communities in which the research participants live may vary on characteristics of ethnicity, culture, and occupation. The COC will have to wait until the sampling design is chosen to suggest specific strategies for community engagement. The COC does believe that some type of advisory or steering committee of community representatives from across the different sampling areas can be created to assist in the engagement of the different communities whose residents will be most affected by the Study.

Members of the expert panel suggested some strategies to facilitate meaningful community participation in the Study. First, the Study could build on partnerships that have already been established. This might involve selection, when possible, of researchers who either have previously undertaken or are currently undertaking research with meaningful community participation in the area of children's health and the environment (see section on RFP selection criteria). Second, choose the sites for the Study as soon as possible and allow 6–12 months for the identification of leaders and establishment of the CABs.

Participation of community representatives is essential for ongoing implementation of the research, including retention of participants and resolving ethical dilemmas that might arise. Community representatives can provide unique perspectives on ethical dilemmas that might arise, especially those concerning cultural or ethnic practices. For example, in a longitudinal

study of environmental triggers for children's asthma[2], the CAB suggested a variety of strategies for retaining children in the study, including:

- Sending them annual birthday cards
- Sending their families newsletters
- Providing annual appreciation parties
- Determining different types of incentives based on the age of the child
- Giving small gifts to the siblings of the child participating to minimize potential disruption to family dynamics caused by the attention focused on the participating child.

Participation of community representatives is important in interpretation and dissemination of findings to the wider community. Community representatives can add insight to the interpretation of the findings given that they are much closer to the lived experience of the participants than are the researchers. Recognizing the value that community representatives can bring in this regard, the expert panel strongly suggested that findings from the Study be shared as soon as is feasible throughout the Study and that communities be allowed to assist with data interpretation. Community representatives can also assist in designing the dissemination of findings to the wider community and ensuring those results are presented in an easily understood and linguistically appropriate fashion.

A challenge of creating CABs is the identification and selection of leaders to represent the community on those boards. In identifying members of a CAB, one needs to focus on both formal and informal community leaders. Although elected officials such as the mayor or city council members might be included, it is just as important to include religious leaders, heads of CBOs and local agencies, as well as other more informal leaders. One way to start the identification process is to do a series of informational interviews with organizational leaders and elected officials in a community. This approach allows one to both explain the Study and seek input and advice about the Study from those already established in the community, and to begin to identify potential representatives for the CAB. At each of these interviews, one should ask whether there are any other persons in to the community who should be interviewed. This technique, called the "snowball method," will generate a list of names. How often a name is mentioned may be one indication of an individual's influence in the community.

Members of the expert panel suggested that once a CAB is established, the researchers at that site should provide training and information about the nature of research to community representatives so that all members can be at the same level of understanding. This training would focus on ethical research versus unethical research and the informed consent process.

Request for Proposal Site Selection Criteria. Achieving the goals of community outreach in the Study depends on the active collaborative of a range of community and research group stakeholders. Community involvement needs to be authentic and to occur early in the design of the Study. When community participants are not routinely involved, the natural concerns of many communities—especially low-income and underrepresented communities—may not be

found within traditional research organizations, advisory bodies, issue-focused interest groups, and commissions established by political, scientific, or business interests.

A Study site may represent a singularly defined community or an assemblage of linked communities that could be described in economic, geographical, or social terms. The panel recommends that sites should:

- Be composed of consortia of well-established institutions
- Be able to affirm and demonstrate a history of productive community engagement as equal partners, an existing collaborative relationship with a university, and an existing community information network
- Have a process in place to continually recruit a cadre of advisory board members from the Study community.

Information to be provided about the relationship between the site and research team includes:

- Description of the research team's experience with similar projects. The overall experience of each team member with culturally diverse communities or community groups should be described. Capabilities and experience in participatory research should be included.
- Description of specific experience with federal, state, and local governments
- Description of the approach to community outreach and communication over the life of the Study, which will fully address the requirements of the request for proposal. Anticipated engagement approaches in response to how communities may be expected to evolve should also be described.
- A work plan for the project that meets the stated deadlines of the partnership
- A proposed methodology for benchmarking performance
- A plan to develop an effective recruitment and retention strategy
- Applicants are asked to demonstrate prior experience with coalition building, enhancing partnerships with health providers and community organizations at a targeted site, and inclusive processes for receiving and analyzing community concerns and input.

The following additional areas of consideration should be used in selecting sites:

- Understanding the purpose of the Study. This refers to the research team's understanding of the Study, the concerns that generated the need for the Study, and the nature and scope of the work involved.
- Soundness of approach. Emphasize the techniques for collecting and analyzing data, sequencing of major steps, and managing the Study.
- Employment. Describe the extent of genuine involvement of minority scientists and students who are connected to the community.

The research team should carefully consider its ability to provide the following to the community(s) in the targeted site:

- Conduct needs assessment examining risks, perception of risks, and communication of risks regarding the study
- Assess community concerns, priorities, cultural values, and goals regarding the study

- Develop culturally appropriate education and communication materials including appropriate media presentations
- Use participatory action research incorporating systematic investigation
- Develop a comprehensive evaluation plan of the engagement strategy
- Prioritize community issues through surveys and focus groups and hold regular public forums
- Identify priority community information needs determined by the community
- Train and employ students in data collection and analysis
- Accommodate and respond to community bias and perceptions using nonconfrontational methods.

There is critical need to consider culturally specific approaches and concepts in order to respond to the needs and values of representative American communities. If sensitivity to cultural variances is absent, communities most in need of information to understand environmental or health risks and their impact are not empowered to prevent future health problems. Principles of community engagement include:

- The optimum public decision-making process concerning children's health is one that is fully understood by the majority participating in and affected by the decisions (parents, caregivers, others).
- Because the community is the final authority, all citizens must have access to the information needed to make informed decisions.
- Research teams are urged to collaborate with community-based, state, local, and regional entities to develop and design the community engagement strategies. Community leaders (formal and informal) and design teams composed of cooperative extension service staff members, community based organizations, university faculty from a variety of disciplines, seasoned clinicians, and a host of volunteer community residents and community development people should be involved at each site.

Postsite Selection Recommended Strategies (*Note- These sections are to be completed later by the Community Outreach and Communications Committee after decisions are made about the study design*)

Once the Study is underway, the value of applying a well-designed comprehensive community outreach strategy cannot be understated.

Special considerations and challenges for the Study include:

- Incentives for the Study must be developmentally and culturally appropriate.
- The issue of interventions that will be developed from the results of the Study must be dealt with up front.
- Inappropriate messages and misinformation about the Study will no doubt arise as the Study progresses.
- A major challenge is change in communities over time.

Definition of Terms

Communities. Nonmonolithic groups with a diversity of characteristics sharing common social, ethnic, economic, and geographical identification with mutual perspectives of the conditions of their lives.

Engagement. The collaboration of communities and their representatives to influence and enhance all phases of the research process.

Community representatives. Persons recruited from the targeted communities with evidenced cultural competence relative to community engagement practices and whose life and or professional experience evidence placing a premium on respecting, valuing, and empowering diverse communities.

Study participants. Individuals and families that have agreed to have health examinations, to provide personal health and lifestyle information along with tissue and body fluid samples, and to have their health and/or living conditions monitored.

Study site. Normally a location or set of locations sharing a common Study parameter linkage that can be described in geographical terms.

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Summary of The National Children's Study Workshop Measuring Racial / Ethnic Discrimination and Racism from a Developmental Perspective

June 21-22, 2004

Introduction

Th National Children's Study Workshop on Measuring Racial / Ethnic Discrimination and Racism from a Developmental Perspective, was initiated by the Health Disparities and Environmental Justice Working Group of the National Children's Study Advisory Council. The goal was to summarize findings concerning how best to measure racism and discrimination in multiple racial / ethnic groups from a longitudinal, epidemiological perspective.

Background

In April, 2002, The Office of Behavioral and Social Sciences Research of the National Institutes of Health convened a meeting of approximately 100 scientists to present scientific evidence of the effects of racial / ethnic bias on physical and mental health (1). In addition to highlighting findings related to associations between racism / discrimination and health outcomes, this comprehensive review of existing literature focused on directions for future research. Among their findings was the conclusion that the majority of empirical research had been done on African Americans and that few studies have systematically addressed how prejudice and discrimination affect other racial / ethnic minority groups (1), and that there is no consensus in the literature as to the optimal measures for capturing exposure to discrimination (2). The latter point is especially relevant for children, where the least amount of research has been done.

A report from the National Research Council, compiled by the Panel on Methods for Assessing Discrimination of the Committee on National Statistics, notes that racial ethnic disparities in health outcomes are well known. The fact that they exist is not proof that racism or discrimination is a causal factor, however it does motivate further investigation into the issue. That report uses a definition of discrimination which goes beyond the legal definition to encompass differential treatment on the basis of race/ ethnicity that disadvantages any racial / ethnic group. The purpose is to include behavior that has negative consequences but may not be unlawful. It categorizes discriminative behavior into four areas: discrimination that is intentional and explicit; discrimination that is subtle, unconscious and automatic; statistical discrimination and profiling; and exclusionary organizational processes. This report further reflects the inherent difficulties in measurement and reiterates the difficulties in defining racial discrimination in a clear way so that credible ways of measuring can be found (3). It says that "For the purpose of understanding and measuring racial discrimination, race should be viewed as a social construct that evolves over time." However, there is no single approach to measuring racial discrimination that allows researchers to address all the important measurement issues or to answer all the questions of interest. Because measurement of race can vary with the method

used, it recommends that whenever possible, multiple methods be employed in the same study so that comparisons can be made across methods..

The purpose of the National Children's Study Workshop on Measuring Racial / Ethnic Discrimination and Racism was to address the issues of measurement from a lifespan perspective, identifying important contextual factors and feasibility issues relevant to a prospective epidemiologic study of children.

Constructs Relevant to Various Ethnic / Racial Populations over Time

The Report from the National Research Council defined *race as a social construct* that evolves over time. This concept was echoed in comments made by participants related to the "concept of self" and racial identity in children, where it was noted that it is important to put racism in a context. The context of change over time is important not only within individuals, i.e., as children grow and develop, but also with respect to societal trends as concepts of race evolve over time. Given the longitudinal nature of the National Children's study, societal changes are likely to occur and should be documented, especially those events that differentially impact racial groups.

From a developmental perspective, attributions such as *racial identity* evolve and change as children grow older. Part of this process stems from learning racial language as they begin to socialize outside the home in day care settings and schools. However, a sense of "place" is also an important issue for racial / ethnic identity. In its most basic context, place simply means a geographic location. But from a social perspective it is also a nexus where social life is initiated and engaged, and involves the values, traditions and history we use for organizing our experiences. A sense of place gives children a sense of security and engagement. Exclusion from social networks can be a form of denying children a sense of place. Children who are caught between the values of two cultures, the dominant white culture to which they do not belong, and their own race / ethnic group, also have more complex issues with respect to a sense of identity than children from the dominant culture. The stability of a child's concept of self may also vary by immigration status. African Americans have a history of discrimination in this country and become aware of it at an early age. It is a stable cultural reality for African Americans in our society. Whereas for immigrants, a child's concept of self and who s/he is may be more inclined to vary over time, depending on their particular culture and the attitudes of Americans towards that culture. A case in point is Muslim children who may be experiencing much more negative feedback about their identity since the events of 9/11 than before.

The example of Muslim children after 9/11 also illustrates that discrimination occurs in a social-cultural context and may manifest differently in different race / ethnic populations. Ways of coping with discrimination also vary by population. It has been demonstrated that passive, forbearance coping reactions are more effective in Southeast Asians with strong attachment to Asian ethnic values than in Koreans (4). In Korean immigrants, active, problem-focused coping was most effective, especially among those who were better acculturated. Although Vietnamese, Chinese and Korean immigrants share the same cultural traditions of Confucianism, Buddhism, and Taoism that would point to a predominance of forbearance coping, Korean immigrants are

much more likely to be affiliated with protestant churches, have more members at higher SES levels and are better acculturated.

It was emphasized throughout the meeting that the association of discrimination with physical and mental health outcomes stems from it cumulative effect. Measuring at one point in time does not provide an adequate conceptualization for meaningful analyses. It is the cumulative effects that result in the primary emotions of anger, sadness and aggression and the cognitive sense of shame, powerlessness or lack of personal control, exclusion and discouragement. The cumulative effects are of several types: generational, i.e., passed from parent to child; longitudinal within domains, such as discrimination within the school system over time from elementary grades through high school; and between domains, i.e., those stemming from different sources such as health care settings, work and school. Thus, data show that overt discrimination is significantly associated with a reduction of positive affect or sense of well-being. Subtle discrimination is not directly associated with a reduction in positive affect but is associated with depression (4).

Implications for measurement: The implication of these data for measurement is that discrimination should be clearly defined and measured in various situations and contexts, from individual situations to institutional settings. It should also be measured multiple times during development and its frequency of occurrence noted along with the multiple settings in which it occurs. However, to understand the impact of discrimination on physical and mental health outcomes, it is also important to measure how well a person copes. Successful coping will increase resilience and reduce, at least somewhat, the negative effects of discrimination. Therefore, it is also important to measure inter-generational experiences because parents pass their expectations with respect to discrimination and their methods of coping on to their children. Since means for successful coping vary across cultures, measurements must be done in a culturally sensitive manner. Emotion and cognition play key roles in coping strategies and should be included as part of this process.

Domains of Measurement

There are several ways of thinking about the domains of measurement from a theoretical perspective. One framework conceptualizes institutionalized vs. personally mediated racism / discrimination, and the extent to which these forms of discrimination have been internalized (5). In this context, institutionalized racism / discrimination is defined as differential access to the goods, services and opportunities of society based on race. Personally mediated racism is defined as prejudice and discrimination at an individual level based on assumptions about the motives, intentions and abilities of others according to race (5). When the discrimination is internalized, the members of the race or ethnic group which has been discriminated against, begin to believe the biases of others and come to believe it, feeling that they are somehow inferior because of their race. One definition of successful coping would be the ability of a child to withstand these assaults on his / her personal sense of self. Another way of conceptualizing race would be the perception of discrimination by the individual, the social distance between races and the racial climate created by relations between race / ethnic groups.

A number of domains and settings were enumerated where racism may occur: residential housing, the physical environment (e.g., neighborhood), schools, school clubs, day care, access to work and work environments, health care, research settings, media & entertainment, immigration, prisons and the criminal justice system, neighborhood surveillance (e.g, by the police), adequacy of nutrition, access to transportation, and internet sources of racism (now quite wide spread). It was emphasized that these various domains contribute to the overall cumulative burden of discrimination. Other contextual measures relate to distribution of local resources (are supermarkets available in the neighborhood or do they have to travel to get to one) and hazards (drugs, alcohol).

Social support systems of all kinds, including affiliation with a faith based community, were identified as important in helping to cope with discrimination. Self-efficacy, hope, optimism, resiliency and personal control were all seen as being influenced by the extent of an individual's exposure to racism and discrimination.

Several participants asked whether the study of discrimination would be limited to race / ethnicity. They said discrimination was also important with respect to gender and sexuality (e.g., heterosexual vs. homosexual), and pointed out that this may be particularly important in instances where family structure in the racial / ethnic groups is confounded by discrimination based on either of these other issues.

Implications for Measurement

Conceptually, there are several ways of gathering data about discrimination in epidemiological studies (6). It can be inferred indirectly at the individual level; measured directly by self-report of discrimination in the individual; and thirdly, at a group level. The latter method involves investigating whether group-level measures of discrimination are associated with population rates of health outcomes..

Data bases: Data bases are one way of inferring data without using self-report. Macro trends in social attitudes can be obtained through sources such as Gallup poles. Geocoding to census tracts can be utilized to obtain information on poverty percentages, median housing values, etc., from which an index of dissimilarity can be created as one measure of institutional discrimination (7). The Home Mortgage Disclosure (HMDA) data base that includes information on loan type, purpose, loan amount requested, applicant income, reason for denial, gender, race and co-applicant characteristics has revealed inequities in lending practices that are objective measures of racial climate and institutionalized racism in specific neighborhoods. Another method of providing context to individual measures of discrimination is the examination of the racial composition of school boards and teacher ratios for representational parity. The general Accounting Office (GAO), the National Academies and over 200 other organizations and individuals are also working together to develop a national indicators system with a broad range of information covering the economy, society, and the environment (8).

Individualized measures of racism: The group expressed consensus that it would be important for any instruments selected for the NCS to be composed of scales that have been tested and shown to have internal consistency, reliability, and / or been validated as predictors of specific

health outcomes. This would enable results from the NCS to be compared to previously published literature and assure that if lack of association were found, it would not be attributable to unreliable measures.

Multiple measures were suggested that are related to the above domains. One such measure that measured discrimination in multiple domains was developed for the longitudinal Coronary Artery Risk Development in Young Adults Study (9,10). This questionnaire measures whether the participant has "ever experienced discrimination, been prevented from doing something or been hassled or made to feel inferior... because of their race or color" in any of seven situations: "at school, getting a job, at work, getting housing, getting medical care, on the street or in a public setting, and from the police or in the courts". This variable can be coded according to how many situations discrimination has been experienced.

A telephone administered perceived racism scale (TPRS) was developed from focus groups of African Americans who evaluated the content and face validity of the questions. Examples of questions include: "Because I am Black, I feel..."; "Whites often assume that blacks work in lower status jobs and therefore, treat them as such" (11). Factor analyses revealed five factors: passive emotions (feelings of powerlessness and hopelessness), active emotions (angry, frustrated, anxious, sad), passive behaviors (don't speak up), internal active behaviors (praying) and external active behaviors (working harder to prove them wrong). Tests of internal consistency indicated good reliability for all scales. This instrument has the advantage of being designed for telephone interviews, which can be important in epidemiologic studies.

Another scale analyzing coping methods in response to discrimination has also demonstrated good internal consistency (12). This measure showed that in Korean immigrants in Toronto, active, problem-focused coping styles were more effective in reducing the impact of discrimination on depression than frequent use of passive, emotion-focused oping, which had a debilitating effect on mental health.

A question measuring race consciousness has also been pilot tested in the 2002 Behavioral risk Factor Surveillance Survey in six states (13). This question asks how often an individual thinks about race and shows large discrepancies between African American and Caucasian participants. Caucasians don't think much about race whereas African American and Hispanic respondents think about it often.

This is not a complete list of the measures discussed at the meeting. Others, along with their references, are being sent to the Program Office. One of these is the Minority Health Survey from the Commonwealth Survey of 2002 (David Takeuchi), another is the MEUSS used by Larry Bobo, another is a measure of social exclusion (Elena Yu), and still another is a measure of "tokenism" b Pamela Jackson.

In the discussion of measurement, it was also suggested that institutionalized racism could be tested in "audit" studies, an example of which would be sending out the same resume to multiple businesses that have advertized for personnel, but varying the names on the resume to resemble distinct ethnic groups or genders. The purpose would be to see if there is any consistent variation in the ranking of the quality of the resume based on these factors. It was

suggested that this may be an appropriate “in depth” study to do with the R01 grant mechanism once the study is established and all centers have been activated.

Measures in Children and the Life Span Perspective

With respect to measuring racism and discrimination in children, two important points were made: measurement in children requires different methods than in adults and although discrimination at all ages is detrimental, there are time points where a child may be more vulnerable to the negativity created by discrimination than at others.

Group discussion indicated that measurement in children cannot be totally separated from that in adults, due to the importance of the mother/child diad. Thus, measures of discrimination in the mothers of the NCS children’s cohort will be important during pregnancy as a measure of stress and its resulting neuroendocrine consequences. In this context, the expectancies of the pregnant mothers for their unborn children will also be important. It was noted that young, pregnant mothers in minority groups, especially in socioeconomically deprived areas, often fear for the survival of their unborn children and that a form of institutionalized racism begins in the clinics, where clinic personnel automatically assume that if a baby was unplanned, it is also unwanted. When mothers are asked whether the baby was planned, if they answer no, there is no follow-up question. Clinic staff automatically assume that it is also unwanted. Simply knowing whether a baby is being born into a welcoming situation or is unwanted might be important for children of all races.

However, measuring discrimination in the mother will also be important after birth, not only as a measure of stress, but as a reflection of the experiences and attitudes that the parent will be conveying to the child. It was noted that the resiliency of the child is to some extent dependent on the resiliency of the mother. Racial coping skills are critical competencies for African American (and probably other minorities) children to have (14). If the mother is resilient and capable of teaching successful coping skills to the child, he or she will have a better chance of faring well despite discrimination.

With respect to domains of measurement in children, it was emphasized that the cumulative trajectory over time is extremely important. The domain of education is an example where discrimination may begin early, taking the form of subtle assumptions about children on the part of teachers, based on the child’s race or socioeconomic status. Whether the trajectory assumes the form of cumulative burden, depends on how often and in what forms discrimination occurs during the progression from elementary through middle school, high school and college.

Objective measures of inequities in education include demographics such as student / teacher ratios in schools, teacher attendance, school resources and physical conditions of schools, as well as standardized test scores and drop out rates. There is also a series of age appropriate measures for primary school children which has been developed to examine school and teacher climate, student racial coping, as well as a child’s self-efficacy and self-concept, that has been developed and validated in the Comer school intervention project (15). The Comer intervention model emphasizes positive, collaborative partnerships among teachers, administrators, families and students (16). The measures developed to assess coping and well-

being include: what I think of school, dialogues about family and friends, a pictorial scale of perceived competence and social acceptance for young children, a self-perception profile for children, a culture-free self-esteem inventory, racial coping measures and a self-efficacy inventory. An important finding of this study is that these concepts are not stable over time but change as the children get older. Experience from this study indicates that effects of discrimination on self-esteem begin as early as 2nd grade.

In younger children, the advantages of projective techniques were also emphasized. These techniques involve having children make up a story related to a picture, having the child respond to a hypothetical situation of another child in a story, or giving the child a camera and telling him or her to go outside of their residence and take a picture, then tell about the picture

Measures of school climate determined from ratings of parents, teachers and students have also been developed and validated in middle school and high school populations (17), showing pervasive racial differences among school staff. Additional measures suggested by workshop participants as being relevant for children include: children's aspirations and the way they change over time; the point at which a child first recognizes race; stereotype threat; homework stress; and white privilege. White privilege is defined as the ability of whites in America to ignore the issue of race because it doesn't affect them. Things that white children take for granted, such as fair treatment in school, are not privileges granted to minority students. However, whites, not confronted with these issues themselves, are often not aware that they exist. Access to "cultural capital" - the extent to which children are given piano and ballet lesson, taken to the theater, and given horse back riding, tennis or golf lessons was also thought to be important. These cultural factors can further serve to separate children along racial lines.

Access to mental health services for children, access to medications (e.g., asthma medication for children who need inhalers is sometimes taken away from the child and required to be kept in the nurse's office), stigmatization about being obese, what a child knows about sexuality (the group realized this may be difficult), and access to physical activity in schools were also considered important.

Community Partnerships

One area that was greatly emphasized in this workshop was the importance of partnering with communities. An observational study such as the NCS must establish an ongoing partnership with the involved communities from the very beginning. The participants and the community need to receive regular feedback concerning study findings and issues related to their communities and individual children (e.g., health related feedback). Other suggested partnerships included the Office of civil Rights, the Department of Housing and the Department of Labor.

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