Statement of Work: August 8 2007

Project Title: Using the “Information Rx” Process to Refer Parents of Newborns with Metabolic Conditions Identified by Screening to the Genetics Home Reference Web Site for Health Information: Feasibility Study in Utah

Cost: $61,945

Project officer: Robert A. Logan, Ph.D., Lister Hill National Center for Biomedical Communications, 8600 Rockville Pike, Building 38A, Room 9s914, Bethesda, MD. 20894; 301 496-1936; logan@nlm.nih.gov

Proposed contractors: Principal investigator - Joyce A. Mitchell, PhD, Chair, Biomedical Informatics, 26 South 2000 East, HSEB Suite 5700, School of Medicine, University of Utah, Salt Lake City, UT. 84112-5750, Joyce.Mitchell@hsc.utah.edu, 801 581-4080;; Principal co-investigators: Denise E. Beaudoin, MD, MSPH, MS, Research Associate, Department of Biomedical Informatics, 26 South 2000 East, HSEB Suite 5700, School of Medicine, University of Utah, Salt Lake City, UT 84112-5750, denise.beaudoin@hsc.utah.edu, 801-581-4080; Nicola Longo, M.D., Ph.D., Director, Metabolic Service, Division of Medical Genetics, Department of Pediatrics, University of Utah, 84112-5750, nicola.longo@hsc.utah.edu, 801-587-7400

Synopsis:

This Statement of Work describes a proposed contract between the National Library of Medicine, the Regional Medical Library at the University of Utah and the Department of Biomedical Informatics, University of Utah.

The focus of the proposed evaluation is the Health Information Rx Program for Newborn Screening and Related Genetic Disorders. This program encourages physicians to direct the parents of newborns with metabolic conditions identified by screening to the Genetics Home Reference (GHR) Web site (http://ghr.nlm.nih.gov).

The proposed work is a pilot project and feasibility study to: a) intervene and provide an information prescription to all parents of Utah newborns who are screened and receive a positive diagnosis of a metabolic condition, b) intervene to urge affected parents to use Genetics Home Reference in order to learn more about their child’s metabolic condition; c) evaluate the impact of both interventions and, d) determine if the pilot is a possible prelude to a regional or national expansion of the proposed interventions.

GHR is designed to provide consumer friendly, comprehensive information about genetic conditions and disorders. GHR is housed in the National Library of Medicine (NLM) and was first established in fiscal year 2005.
The Health Information Rx Program for Newborn Screening and Related Genetic Disorders, which is broader than the pilot project discussed in this Statement of Work, is called the GHR Information Rx project for the remainder of this document.

Overall, the GHR Information Rx project seeks to improve newborn health by providing an authoritative, user-friendly, commercial-free information resource about genetics to the parents, families and caregivers of affected newborns. The pilot project’s triggering event is the diagnosis of a metabolic condition detected by newborn screening.

Program partners for the existing GHR Information Rx project include: the National Institute of Child Health and Human Development, the American Academy of Pediatrics, the American Academy of Family Physicians, the American College of Obstetrics and Gynecology, the American College of Medical Genetics, the School of Medicine at the University of Utah, the Utah Department of Health, and the NLM.

The GHR Information Rx project joins other information prescription efforts by the NLM that encourage health care providers to prescribe a Web-based health information resource within routine patient-provider encounters. NLM’s broader Information Rx programs are co-sponsored by NLM’s Office of the Director, the Office of Communication and Public Liaison, and the Office of Health Information Programs Development.

There have been two previous evaluations of GHR conducted by the Cognitive Science Branch of the Lister Hill National Center for Biomedical Communications, which oversees the contract for this proposed assessment. We envision the proposed evaluation as a pilot project within the context of the larger GHR Information Rx project.

**Description of Services:**

Professional services are required over a 12-month period for implementation (phase 1), data collection, and analysis for the GHR Information Rx project.

The requested budget covers program origination, implementation and evaluation of clients.

**Background:**

The purpose of the proposed evaluation is to assess a patient education intervention and a consumer health information resource from the perspective of affected parents. In addition, the evaluation of GHR Information Rx will contribute to consumer health informatics research as parents who visit GHR will provide valuable feedback about the information available on the site.

Ultimately, the evaluation will determine if the GHR Information Rx project is a model that can be used in other states. The evaluation also should help NLM improve its information prescription efforts with consumers and provide feedback to the NLM team that produces GHR.
The broad rationale for the evaluation of GHR Information Rx is general interest about how families assess the dual intervention of receiving an information prescription along with a recommendation to visit a specific Web site or consumer health information resource.

There is evidence that parents of children who are referred for a possible genetic diagnosis experience stress, fear and anxiety, and need professional input to help them access additional information, explanations, and psychological support. The GHR Information Rx pilot project will attempt to reduce parental anxiety by providing reliable and consumer friendly information about their child’s condition. The proposed evaluation will assess whether receiving an information prescription and visiting a health information resource prompts constructive parental action, such as discussing the information with their child’s pediatrician, contacting a support group, or looking for additional health information.

The results should improve both the information prescription process and provide insights about GHR as a health information resource among one of the Web site’s intended audiences. The evaluation also should suggest if the GHR Information Rx project is a model that can be used in other states.

Finally, the evaluation will supplement previous findings about information prescription interventions by other divisions within NLM and previous consumer feedback about GHR. These insights will help NLM improve both its information prescription efforts with health care providers and consumers and provide feedback to enhance GHR to the NLM team that produces the Web site.

**Proposed Methods**

New data will be collected in a prospective study of GHR Information Rx. This will be an event triggered, feasibility study of the universe of Utah parents of newborns with a positive screening test for a metabolic condition. The parents of newborns who test positive for a metabolic condition identified by newborn screening will receive an information prescription to visit the GHR site in order to learn more about their child’s condition. The specific metabolic conditions recommended by the contractor to trigger parent participation are listed in Appendix C.

Participants will receive two survey instruments. An initial paper and pencil survey instrument will be derived from an NLM developed questionnaire that was previously used by the American College of Physicians to assess how physicians and patients evaluated an information prescription program. The survey will be pilot tested by a convenience sample of parents who are ineligible for the study (e.g., do not have a child with one of the specified conditions), revised as needed and finalized prior to implementation. At least one patient/caregiver will be asked to complete the initial survey at the time of the study enrollment.
A follow-up survey will provide an evaluation of the information prescription process as well as information seeking habits, such as whether respondents used other health information resources to learn more about their child’s condition. The follow-up survey will be administered online and will be accessible from a ‘mock’ GHR site. Families without access to the Internet from home will be directed to public libraries or offered assistance from librarians at the Eccles Health Sciences Library, University of Utah.

Parent will receive a ‘reminder letter’ three weeks from the date of study enrollment reminding them to visit the GHR site. Parents will receive a second ‘reminder letter’ (containing the URL needed to access the site and online survey) approximately six weeks from the date of study enrollment requesting them to visit the GHR site and complete the online survey. Three attempts (by telephone and/or email) will be made to contact study participants who have not completed the follow-up survey at the end of the six-week period to further encourage compliance.

Outcome variables in the questionnaire include: user exposure to the intervention, user understanding of the genetic disorder and medical condition, health information seeking behaviors, patient-provider interpersonal communication, user reaction to the Information Rx intervention, user satisfaction with GHR compared to other health informatics services, user assessment of the quality and trustworthiness of GHR, user evaluation of specific GHR features, pre-post intervention exposure outcomes, continued use of GHR, use of other health information resources, Internet use, and demographic information.

A discussion of the clearances, informed consent and other data collection and privacy issues is provided under IRB clearance below.

**Proposed Work:**

The proposed work will be conducted at the Health Sciences Center at the University of Utah. All three proposed contractors are faculty at the Health Sciences Center at the University of Utah.

Dr. Mitchell, the project director, will provide administrative oversight of the project. Dr. Beaudoin, who is a research associate, will primarily assist with project planning, origination, data collection, data analysis and dissemination. Dr. Longo, who is a project associate, will primarily assist with identifying patients to participate in the study as well as ensuring patient compliance to the study’s proposed interventions.

The contractor has secured access to the universe of Utah parent of newborns with a positive screening test for a metabolic condition.

The NLM project officer will be consulted regarding all aspects of the research instrument, its implementation, analysis and dissemination. The contractor will be asked to provide bi-weekly short reports and monthly updates of the contracted research. The collaboration between the Project Officer and the contractor is enhanced by two pre-existing conditions: 1) the surveys used in the evaluation are derived from instruments previously used by the Project Officer and the National Library of Medicine in other
information prescription projects, and 2) there is an established working relationship between the Project Officer and the Contractor.

The study will be initiated as soon as the project has been approved and Express Evaluation Funds are awarded. The project timeline is one year. The project’s timeline with assigned responsibilities is in Appendix A, which is attached.

In Utah, parental notification of a positive test result is coordinated by staff in the Newborn Screening Program at the Utah Department of Health (UDOH). The UDOH first contacts the affected family’s primary care physician. The primary care physician then either informs the parents of the positive test result or requests the UDOH to do so. The parents are then referred to the metabolic clinic, directed by Nicola Longo, M.D., Ph.D., at the University of Utah. Dr. Longo sees every child in Utah who is diagnosed with a metabolic condition. Thus, the entire universe of metabolic conditions rather than a sample of Utah’s affected population will potentially be surveyed.

Dr. Longo provides information about the metabolic condition to parents once he is certain of the diagnosis (usually at the first clinic visit but sometimes at a second visit). He also gives parents two copies of a book chapter relevant to their child’s condition, instructing them to keep one copy and give the other to their child’s primary care physician.

On average, Dr. Longo sees between 10 and 15 patients at the metabolic clinic each Monday (both newly diagnosed patients and those already diagnosed). According to Dr. Longo, five patients could potentially be enrolled into the study each week for a total of 60 patients during a three-month study period (conservative estimate). This could result in data collection from 60 to 12 parents/caregivers (depending upon whether one of both parents/caregivers want to participate in the study). As children with hearing abnormalities, hemoglobinopathies, and endocrine disorders are diagnosed and treated by various other providers in the state, this pilot project would focus on the metabolic conditions diagnosed by Dr. Longo (please see attached list of conditions).

Parents will be enrolled in the pilot study at the clinic. Dr. Longo will give all parents an ‘Information Rx’ to visit the GHR site when he informs them of their child’s diagnosis. Upon completion of the consultation with Dr. Longo, Dr. Beaudoin will speak briefly with parents to describe the study, invite participation, and obtain informed consent.

IRB clearance

The proposal will be reviewed and approved by the University of Utah Institutional Review Board prior to the study’s implementation and all other required clearance processes. The project is exempt from OMB clearance since patient clinical data will be collected. All data collection, gathering and reporting will be done exclusively by the proposed contractor at the University of Utah. All participants will qualify for the study only after signing an informed consent form and authorization of releasing aggregate research findings, which will be carefully explained prior to requesting a signature from
each respondent. All personal demographic, clinical and other data gathered from survey instruments and the respondents will be kept exclusively and in confidence by the proposed contractors at the University of Utah. The National Library of Medicine will not request or receive any personal demographic or clinical data from the respondents. As a result, any reports of findings will be based on aggregate data and will not disclose any personal demographic or clinical information. All confidential information will be kept on a password protected computer and/or in a locked file cabinet as appropriate. Data will be collected on an ongoing basis, entered into a database, and checked for accuracy prior to analysis. Descriptive and analytical statistics, including determination of general frequency responses to all survey items, will be used to assess responses to the study questions.

Dissemination of findings

A summary report of the GHR Information Rx evaluation results, based only on statistical aggregates, will be provided to the NLM. The authors also will prepare a brief, plain language report that explains the study’s aggregate findings. Aggregate results from the study will be made available to Utah health care providers and the general public. It is expected that the findings of the study will be of particular interest to pediatricians who, upon diagnosing these conditions, seek to refer patients to reliable resources for supplemental health information. The results may be used to provide feedback to the developers of GHR and determine the feasibility of a regional or national study GHR Information Rx project.

Required tasks:

The following tasks are to be completed as outlined. Also, please see the project timeline in Appendix A.

Task description:

The professional services to be performed consist of the delivery of hours of professional services in support of the following tasks:

Phase 1

Subtask 1. Consult with the Project Officer regarding the instruments to evaluate the project. Apply for funding from NIH’s Express Evaluation Award. Complete survey instruments.

Subtask 2. Obtain Institutional Review Board (IRB) approval at the Health Sciences Center, University of Utah.

Subtask 3. Consult with Dr. Longo on the appropriate time to initiate the study.

Subtask 4. Prepare materials to explain the study to future participants
Phase 2

Subtask 5. Initiate the study – follow protocol for pre-post implementation of survey instrument outlined in the Statement of Work

Phase 3

Subtask 6. Prepare comprehensive data entry of all results within a statistical package that is compatible with LHNCBC’s existing statistical software, SPSS 16.

Subtask 7. Perform data analysis of all results within a statistical package that is compatible with LHNCBC’s existing statistical software, SPSS 16.

Subtask 8. Prepare an executive summary of key findings and write manuscripts for professional dissemination of the results. These manuscripts will be submitted to the Project Officer for review.

Deliverables:

Assuming an Express Evaluation Award is received, partial payments will be authorized to the contractors through LHNCBC. The deliverables under this statement of work shall be provided by the Contractor according to the following schedule.

Phase 1

Deliverable 1: 1 month after contract begins upon completion of survey instruments as proposed in Appendix A. PARTIAL PAYMENT: $20,648 to Health Sciences Center, University of Utah.

Phase 2

Deliverable 2: 4 months after contract begins after initiation of project’s proposed second survey as proposed in Appendix A. PARTIAL PAYMENT: $20,648 to Health Sciences Center, University of Utah.

Phase 3

Deliverable 3: 11 months after contract begins after draft report of project is received (as proposed in timeline below in Appendix A). PARTIAL PAYMENT: $20,648 to Health Sciences Center, University of Utah.

Budget:

The project budget is in Appendix B, which is attached.
The project officer and contractors recently applied for an NIH Express Evaluation Award to cover the anticipated $61,945 budget for this project.

Notice of Government Unlimited Rights to Work First Produced Under this Contract

Government rights to work first produced under this contract are established by Federal law including, but not limited to, this specific reference: FAR 42.227-14, Rights in Data – General, (b)(1).

Requirement to Notify Government of Proprietary Work Dependencies

Offerors are required to notify the Government in writing of any dependencies of the deliverables under this contract on proprietary, copyrighted, or patented work that potentially inhibits, restricts, or requires permission for the dissemination of the deliverables to the public, other governmental agencies or research groups, or any other parties whatsoever.

Sole Source Justification:

There are six justifications for a sole source award. First, a feasibility study of the Genetics Home Reference’s Information Rx program is a current priority within the National Library of Medicine. The sooner a feasibility study is conducted, the faster decisions can be made about expanding the project.

Second, the contractors provide an exceptionally well-organized and planned project to assess the feasibility of GHR Info Rx program at an opportune time and at a modest budget.

Third, the project features a modest budget (under $62,000) solely because the Principle Investigator or Project Director at the University of Utah is donating her time for this project. This time donation alone is a compelling rationale for a sole source award and explains why the budget for a feasibility study is under $62,000. It is highly unlikely that other potential contractors will be so committed to a feasibility study and the success of GHR’s Information Rx program that they will voluntarily waive their time and demonstrate similar good faith. Hence, the costs to NLM are significantly reduced by working with the proposed contractors.

Fourth, the three proposed contractors are uniquely qualified to do this project.

Fifth, the relationships among the contractors and the working relationship among the contractors and the National Library of Medicine, including officials within the Lister Hill National Center for Biomedical Communications, is highly unlikely to be duplicated in other states, or settings. Dr. Joyce Mitchell, who is the project’s principal investigator, founded Genetics Home Reference in 2003 and remains a consultant to GHR. Dr. Beaudoin, who is a co-principal investigator on this project, was an informatics training fellow at the Lister Hill National Center for Biomedical Communications in 2003. Dr. Longo, who is a co-principal investigator, is a colleague of Dr. Mitchell and Dr.
Dr. Mitchell will oversee the project; Dr. Beaudoin’s and Dr. Longo’s roles and responsibilities are explained previously. As a result, there is an established working relationship between the contractor and LHNCBC as well as a track record of conducting GHR and information prescription evaluations between the Contractors and the Project Officer.

Dr. Mitchell founded Genetics Home Reference, initiated the Genetics Home Reference information prescription project and is chair of the department of biomedical informatics at the Health Sciences Center, University of Utah. Dr. Beaudoin and Dr. Longo are both physicians at the Health Sciences Center, University of Utah. Dr. Beaudoin is an experienced researcher in the field of consumer health informatics. Dr. Longo, as director of the University of Utah Metabolic Clinic, sees every child in Utah who is diagnosed with a metabolic condition and consults with the child’s parents or guardians.

Sixth, the setting at the University of Utah provides an ideal locale for the proposed pilot project since: a) the three investigators are professional colleagues, b) Utah is one of the few U.S. states where the screening and diagnosis and disclosure of newborn metabolic conditions to parents are administered by one physician-gatekeeper and c) the Regional Medical Library that will be the funding mechanism for this proposal is located at the University of Utah. This arrangement ensures the successful contact of the study’s respondents and encourages respondent compliance with the interventions and project evaluation.

### Appendix A - Project timeline

<table>
<thead>
<tr>
<th>Tasks</th>
<th>Month</th>
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<tbody>
<tr>
<td>1</td>
<td>Receive Approval for Evaluation Set-Aside Funds and Inform Project Partners</td>
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<tr>
<td>2</td>
<td>Develop Data Collection Forms and Create Database</td>
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<td>2.1</td>
<td>Revise survey instruments</td>
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<tr>
<td>2.2</td>
<td>Develop study “packet” containing cover letter, study description, consent form, initial questionnaire</td>
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<tr>
<td>2.3</td>
<td>Develop procedures for data collection</td>
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<tr>
<td>2.4</td>
<td>Create database to store study data</td>
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<tr>
<td>3</td>
<td>Obtain Approval to Conduct Study</td>
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<tr>
<td>3.1</td>
<td>Complete new study forms and submit to University of Utah’s Institutional Review Board</td>
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<tr>
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<th>Pretest Survey</th>
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<tr>
<td>4.1</td>
<td>Locate convenience sample of parents to pretest survey instruments</td>
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<tr>
<td>4.2</td>
<td>Conduct pretest of survey instruments</td>
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<tr>
<td>4.3</td>
<td>Implement improvements in survey instruments based on pretest feedback</td>
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<tr>
<td>4.4</td>
<td>Move follow-up survey instrument to online environment</td>
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<tr>
<th></th>
<th>Implement Study</th>
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<tr>
<td>5.1</td>
<td>Determine logistics of on-site enrollment of study participants</td>
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<tr>
<td>5.2</td>
<td>Distribute “Info Rx” materials to Dr. Longo and office staff</td>
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<tr>
<td>5.3</td>
<td>Enable link to online survey</td>
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<thead>
<tr>
<th></th>
<th>Collect and Analyze Data</th>
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<tbody>
<tr>
<td>6.1</td>
<td>Initiate data collection from parents (initial survey)</td>
</tr>
<tr>
<td>6.2</td>
<td>Conduct online follow-up survey</td>
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<tr>
<td>6.3</td>
<td>Perform ongoing data entry</td>
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<tr>
<td>6.4</td>
<td>Perform ongoing quality control of data</td>
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<tr>
<td>6.5</td>
<td>Conduct data analysis</td>
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<thead>
<tr>
<th></th>
<th>Synthesize/Disseminate Study Findings</th>
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<tr>
<td>7.1</td>
<td>Submit draft report to NLM</td>
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<tr>
<td>7.2</td>
<td>Revise draft report</td>
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<tr>
<td>7.3</td>
<td>Submit final report</td>
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<tr>
<td>7.4</td>
<td>Post final report online</td>
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<tr>
<td>7.5</td>
<td>Prepare manuscript</td>
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Appendix B – Budget

CONTRACTOR COSTS

DIRECT LABOR COSTS

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<tr>
<th>Labor Category</th>
<th>Hourly Rate</th>
<th>Hours</th>
<th>Amount</th>
<th>Total</th>
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<td>$6,072</td>
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SUBTOTAL DIRECT LABOR COSTS $32,879

INDIRECT LABOR COSTS

Fringe Benefits Research Associate (10% of Direct Labor Costs) Project Associate (33% of Direct Labor costs) $2681; $2004

OTHER DIRECT COSTS (ODC)

Miscellaneous Supplies/Services (based on a potential sample size of 120)
- Copy cost of study packet: cover letter, study description, initial questionnaire, consent form (originals and parent copies) $192
- Postage for return of initial survey (from parents who are unable to complete the survey at the clinic) $50
- Copy cost of “reminder” letters at 3 and 6 weeks $24
- Postage for “reminder” letters at 3 and 6 weeks $100
- Envelopes (for return mailing of initial survey and “reminder” letter) $47
- Printer ink cartridge $60
- Long-distance telephone charges for follow-up of non-responders $100
- SPSS software $300
- Report printing costs $70
- Publication page charges $350

SUBTOTAL ODC $1,355

SUBTOTAL DIRECT LABOR, INDIRECT LABOR, ODC $38,919

TRAVEL COSTS

Project Coordination Meetings/Travel (coordinated by NIH)
- Air fare from Salt Lake City to D.C. (2 people x $800) $1,600
- Per diem costs per meeting (2 people x $64 per diem x 2 days) $256
- Hotel (2 people x $165/night x 2 nights) $660

SUBTOTAL TRAVEL COSTS (for 1 meeting) $2,516 $2,516

SUBTOTAL CONTRACTOR COSTS $41,435 $41,435

FACILITIES AND ADMINISTRATIVE COSTS*** $20,510 $20,510

TOTAL ESTIMATED COST $61,945

* Joyce Mitchell, as Project Director and Principal Investigator, will provide project oversight but will not charge labor costs

*** 49.5% of contractor costs charged by the University of Utah for participation in federal contracts
Appendix C - List of metabolic conditions proposed for study:

- Isovaleric acidemia (IVA)
- Glutaric acidemia type I (GA I)
- 3-OH 3-methyl glutaric aciduria (HMG)
- Multiple carboxylase deficiency (MCD)
- Methylmalonic acidemia (mutase deficiency) (MUT)
- 3-Methylcrotonyl-CoA carboxylase deficiency (3MCC)
- Methylmalonic acidemia (Cbl A,B)
- Propionic acidemia (PROP)
- B-Ketothiolase deficiency (BKT)
- Medium-chain acyl-CoA dehydrogenase deficiency (MCAD)
- Very long-chain acyl-CoA dehydrogenase deficiency (VLCAD)
- Long-chain 3-OH acyl-CoA dehydrogenase deficiency (LCHAD)
- Trifunctional protein deficiency (TFP)
- Carnitine uptake defect (CUD)
- Phenylketonuria (PKU)
- Maple syrup disease (MSUD)
- Homocystinuria (due to CBS deficiency) (HCY)
- Citrullinemia (CIT)
- Argininosuccinic acidemia (ASA)
- Tyrosinemia type I (TYR I)
- Biotinidase deficiency (BIOT)
- Classical galactosemia (GALT)

References:


3. Personal communication with Jane Fun, Genetic Home Reference staff, on March 19, 2007.