

Valuation of Developmental and Reproductive Effects: A Scoping Study

Matt Cannon, Raymond Kopp, and Alan Krupnick
Resources for the Future

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Executive Summary

While environmental factors are known to have a wide range of impacts on human health, recent attention has focused on the harmful effects of exposure to certain pollutants on the human reproductive system and development of the fetus. These effects include reduced fertility, low birth weight, and genetic defects (e.g., skeletal malformations). Although the dose-response relationship between exposure to such pollutants and their effects on the human reproductive system and the fetus has not been established, the studies suggest that these effects may be clinically significant and pervasive, leading to potentially serious public health problems in the developed world.

If the link between these endocrine disruptors and the human reproductive system is definitively established, the policy response will depend on the extent of the biological consequences of exposure. In addition, the choice of appropriate policies will also depend on both the costs imposed on society through activities necessary to reduce exposure and, more important to the present study, the benefits to social welfare obtained if the resulting biological improvements are realized. Thus, it is necessary to determine the value society places on avoiding the reproductive and *in utero* problems that might be caused by these types of pollutants. Such valuation information will be useful to policy discussions of exposure mitigation where the benefits of that mitigation will inevitably (and justifiably) be compared to the cost.

Resources for the Future has completed a scoping study of the value society places on avoiding the above noted reproductive and *in utero* problems. This study has two major goals:

- provide a state of the art assessment of the literature and data sources that may be useful for addressing or estimating individual and social economic values for improving fertility, for reducing incidence of low birth weight babies, and for reducing the rate of fetal abnormalities.
- recommend methods to estimate such values that could be used in subsequent, detailed studies of the benefits of avoiding reproductive and fetal development risks.

The findings of the study include:

1. There are very few existing studies that have used stated preference techniques to directly estimate the benefits associated with a reduction in the endpoints under consideration. Existing analyses are limited to a handful of studies estimating willingness to pay for the *in vitro* fertilization infertility treatment. Furthermore, shortcomings in these few studies limit their application to broader analysis.
2. Elements of the infertility treatment process, such as cost differences across couples due to differences in insurance coverage and the existence of adoption as an alternative to treatment, could possibly provide the means to use revealed preference techniques

to estimate the value placed on the ability to produce a child. However, difficulties such as a lack of existing data and the adverse selection problem associated with medical insurance preclude the implementation of these techniques.

3. Cost-effectiveness analyses have been performed on the in vitro fertilization treatment process in order to determine the efficacy of a rapidly expanding technique that has not been carefully evaluated. Information included in these studies might be used in an estimation of demand for treatment. For example, charges for a single episode of in vitro fertilization have been estimated to be roughly \$8,000. Similar cost information does not exist for other, more common infertility treatment procedures.
4. Existing studies related to low birth weight and birth defects that might be used in value estimation are limited to cost of illness analyses. The incremental cost of low birth weight from birth to age fifteen was estimated to exceed \$5.4 billion in 1988 (Lewit et al., 1995). Similarly, the cost of 17 major birth defects and cerebral palsy was estimated to be \$8 billion in 1992 (Waitzman et al., 1995). These studies, however, only provide a lower bound estimate of the true costs associated with the incidence of these endpoints.
5. Information regarding the prevalence of infertility is mainly limited to one series of government surveys, the National Survey of Family Growth. Infertility treatment centers maintain a registry containing information on the utilization of several infertility treatment techniques. Utilization information is not collected for all infertility treatments.
6. Data related to low birth weight and birth defects can be obtained from the vital statistics system, periodic surveys, and monitoring programs. Relevant information includes incidence of these endpoints and factors influencing the likelihood of the occurrence of the endpoints.
7. Estimation of the value of a reduction in the incidence of infertility may be derived from the infertility treatment market. Specifically, treatment demand curves and the corresponding welfare measures can be estimated. The infertility treatment process is associated with nonpecuniary costs and subjective perceptions concerning the efficacy of treatment. As a result, it is recommended that future research employ both stated and revealed preference information in the estimation process.
8. Existing cost of illness analyses related to the costs associated with low birth weight and birth defects do not reflect the total costs associated with the occurrence of these endpoints. Total costs include the costs of averting and mitigating actions, the cost of lost time and the disutility associated with the endpoints.
9. Characteristics associated with the occurrence of low birth weight and birth defects distinguish these endpoints from other conditions examined in existing models of morbidity. For example, the disutility associated with the endpoints is incurred both by the affected child and by the parents of the child. As a result, it is necessary to develop a formal economic model that explains the behavior of family members and the costs associated with these endpoints.

10. Estimates of the true benefits associated with a reduction of the incidence of low birth weight or birth defects can be obtained through willingness to pay estimates or estimates inferred from actions taken by parents that affect the likelihood of the occurrence of one of these endpoints.

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Chapter 1

Study Objectives and the Concept of Economic Value

1. Introduction

While environmental factors are known to have a wide range of impacts on human health, recent attention has focused on the harmful effects of exposure to certain pollutants on the human reproductive system and development of the fetus. These effects include reduced fertility, low birth weight, and genetic defects (e.g., skeletal malformations). For instance, within the past several years, studies published in medical journals indicated a decline in the male sperm count over time (Carlsen et al., 1992; Auger et al., 1995). Some studies have identified certain chemicals -- termed "endocrine disruptors" -- as a culprit (Colborn et al., 1996). Although the dose-response relationship between exposure to such pollutants and their effects on the human reproductive system and the fetus has not been established, the studies suggest that these effects may be clinically significant and pervasive, leading to potentially serious public health problems in the developed world.

If the link between these endocrine disruptors and the human reproductive system is definitively established, the policy response will depend on the extent of the biological consequences of exposure. In addition, the choice of appropriate policies will also depend on both the costs imposed on society through activities necessary to reduce exposure and, more important to the present study, the benefits to social welfare obtained if the resulting biological improvements are realized. Thus, it is necessary to determine the value society places on avoiding the reproductive and *in utero* problems that might be caused by these types of pollutants. Such valuation information will be useful to policy discussions of exposure mitigation where the benefits of that mitigation will inevitably (and justifiably) be compared to the cost.

Resources for the Future has completed a scoping study of the value society places on avoiding the above noted reproductive and *in utero* problems. This study has two major goals:

- provide a state of the art assessment of the literature and data sources that may be useful for addressing or estimating individual and social economic values for improving fertility, for reducing incidence of low birth weight babies, and for reducing the rate of fetal abnormalities, and
- recommend methods to estimate such values that could be used in subsequent, detailed studies of the benefits of avoiding reproductive and fetal development risks.

It is important to note that this report does not consider the credibility of the literature on the dose-response relationships between certain chemicals and the health endpoints of

interest. We simply presume that such relationships exist and use the literature to define the endpoints with enough precision to develop approaches for their valuation.

2. Valuation Methods

In this section we present a background discussion of the techniques of economic valuation as applied to market and non-market "commodities." We also include a discussion of techniques used to express relative preferences for health outcomes that do *not* involve monetization. These techniques are collectively termed "quality of life" indicators and they are used extensively in estimates of the cost-effectiveness of medical interventions. The background includes a careful definition of terms, such as *value*, and, because of the controversial nature of valuing health and reproductive effects, a justification for expressing preferences for these types of "nonmarket commodities" in monetary terms.

2.A. The Basics

In estimating the benefits of environmental improvements we are concerned about the benefits to society -- what economists term social welfare. The concept of social welfare is meant to be a single measure that captures as many as possible of the important features of well-being that might be affected by a policy involving environmental improvement. For example, if a range of policies under consideration only affected one thing, GDP per capita, and we could state that all other things equal, higher GDP per capita led to greater well-being, then we could base our measure of social welfare solely on GDP per capita. In fact, economists have long recognized that, in general, GDP per capita is not a reliable measure of either individual or social well-being, because market values do not encompass all important economic values (including the importance of environmental protection, among other examples), and because markets that do exist may suffer from distortions that mask underlying economic values (as in the exercise of market power by a monopoly). Generally, it is clear that many important aspects of well-being could be left out by a simple, single attribute measure of welfare. However, a measure of social welfare need not capture all aspects of well-being to be useful for decision making.

Constructing the measure of social welfare used in a cost-benefit analysis (CBA) can in principle be divided into two steps.¹ In the first step one attempts to develop measures of well-being for individuals within a society. In the second step one aggregates the measure of individual welfare to form a measure of aggregate social welfare. The individual measures are subject to two critical concerns: the appropriateness of the single measure chosen as a valid measure of an individual's well-being, and the problems one faces when attempting to quantify the components of the measure. The appropriateness of the aggregate measure depends on both the appropriateness of the individual measures and the aggregation of those measures.

¹ Formally, the measure of social welfare used in CBA is an index. That is, it is a mathematical aggregation of numerous components to form a single numeric value.

Individual measures of well-being are premised on a fundamental economic assumption. That assumption states that the satisfaction of individual preferences gives rise to individual well-being. Economists take this assumption as a matter of faith and it underlies most if not all of economic theory. Others reject this assumption outright. At its base, the assumption means that individuals know what is good for them (i.e., what will enhance their well-being), their preferences for actions and outcomes reflect this knowledge, and they act in a manner consistent with these preferences in a desire to increase their well-being. The validity of the "preference satisfaction" assumption has been debated since Bentham and will continue to be debated. There is nothing we can add to the debate but to simply note the crucial importance of the assumption to the intellectual foundation of CBA.²

If one accepts the preference satisfaction assumption, then we can look to the actions of individuals as guides to their well-being. For example, if we see an individual exchanging \$3 for a six-pack of beer we can state that the exchange made the individual better off (i.e., increased the individual's well-being), but how much better off? The answer to that question brings us to the concept of *economic value*.

To economists, the term *value* has a very specific meaning that we hereafter refer to in this paper as *economic value*.³ The most important features of economic value are that it is a theoretical construct and that monetary measures of economic value are inferred by analysts from the actions that individuals make in accordance with their preferences. Economic value cannot exist independent of an action, in particular, a type of action that requires individuals to make a *choice* where something is given up and something is gained.

For microeconomists, the study of choice allows economic values to be defined and quantified.⁴ A choice implies that an individual is confronted with a selection of alternatives and the consideration of the alternatives by the individual defines a tradeoff.

² If one wishes to delve into the debate regarding preference satisfaction one can begin with the exchange between Sagoff and Kopp in Spring 1993 issue of *Resources*. More strictly philosophical discussions can be found in Williams (1985) and Scanlon (1991).

³ The term value as used by economists causes a great deal of confusion. For example, if someone asked you what are your "values" you probably would not respond by saying \$2.00 for a Big Mac or \$30 for a round of golf at a public course. Rather, when asked about your values, you might say things like honesty or hard work. Similarly, if someone asked you what value do you place on the environment, you might say the need to preserve it for future generations, or you might mention your commitment to environmental stewardship and conservation. You would probably not say \$32 per day to view Bald Eagles along the California Coast.

⁴ One may be inclined to say that the analyst estimates the economic value rather than constructs the value. In fact, the economic literature routinely refers to "value estimates." However, we use the verb *constructs* to underscore the notion that economic value does not exist in a free standing fashion amenable to empirical measurement. Rather, economic value can only be measured with reference to a choice, and the characteristics of that choice largely determine the measured value.

Contemporary economic theory of individual behavior, based on the assumption of preference satisfaction, suggests that when confronted by choices, what is chosen must be at least as desirable, from the perspective of the individual making the choice, as the alternatives that were not chosen. For example, if an individual chooses to relinquish three apples for a peach, an observing analyst can state that under the circumstances of the choice (perhaps known in their entirety only to that individual), the economic value to that individual of the peach is at least three apples. If the choice were giving up \$1 for the peach, and the individual chose the peach, then the analyst would conclude that the value of the peach to that person was at least \$1.⁵

Returning to the problems of measuring changes in individual well-being, suppose a policy is being considered that would lower the price of peaches by 25% and have no other consequences. From the perspective of our individual who is willing to pay at least a dollar for a peach, the policy enables him or her to pay only \$0.75. The difference between the amount given up and the economic value of the peach is a monetary measure of the individual's increased well-being -- in this case \$0.25.⁶

Below we discuss approaches by which the analyst attempts to place economic values on "commodities" that do and do not exchange on markets (e.g., reductions in the risk of exposures to health impairing air pollutants). What we wish to note here is that quantifying the economic value of "commodities" that are affected by policies, and then determining how the policy affects the trade-off individuals make regarding those "commodities," permits the economist to quantify the change in an individual's well-being brought about by the policy.

A crucial step in CBA is the aggregation from measures of individual welfare to a measure of social welfare. In the most common applications of CBA, the aggregation of individual welfare treats all individuals anonymously. That is, no person's welfare is weighted more heavily in the aggregation than anyone else's. The changes in all individuals' welfare are simply totaled up. If a policy increases the welfare of rich people and decreases the welfare of poor people, but the rich peoples' gain outweighs the poor

⁵ To monetize economic value, the foregone alternative (defined by an individual's choice within a specified trade-off) must be expressed in dollars. Unfortunately, this monetization has sometimes created misconceptions. For example, it has been suggested that economic values are confined to prices observed in markets. These misconceptions arise because many people commonly think of the monetary measure of economic value as a *price*: if a widget sells for \$6 in a market, then \$6 must be its value. This view is misleading, however. When a person buys a widget the analyst only learns that it is worth at least \$6 to the buyer. He or she might be willing to pay much more than \$6 if necessary to get the widget. Markets do offer opportunities for people to make choices, but it is these choices and the circumstances relevant to them, that permit construction of the underlying economic values, not the existence of markets and market prices *per se*.

⁶ Policies rarely affect only one good or one price. Most often they affect many goods and many prices. But if we knew the economic value of all the goods affected by the policy and the effects on those goods of the policy, we could aggregate the monetary measures of well-being gains or losses across all the affected goods to capture the full impact of the policy on individual well-being.

peoples' loss, the anonymous aggregation rule would label this change an improvement in aggregate social welfare.⁷

With respect to measuring welfare gains from health improvements, there is an important additional step. Measures of individual welfare will not capture medical costs paid by insurance companies. Thus, in moving from an individual to a social measure of a welfare change, the difference between what the individual pays for medical care and what that care costs society needs to be captured in the estimate of social welfare.

2.B. Approaches to Measuring Values

The techniques for measuring welfare improvements fall into two general categories: stated preference and revealed preference. The stated preference technique involves asking people questions in surveys to elicit, either directly or indirectly, estimates of the willingness-to-pay (WTP) for the improvement in question. The revealed preference technique involves examining behavior, either in the marketplace or elsewhere, to discern WTP. Examples of the former approaches are contingent valuation (CV) methods, which are structured surveys meant to elicit preferences in monetary terms when confronted with a choice, and conjoint analysis, an approach used extensively in marketing to elicit preferences for particular combinations of product attributes.

There are a wide variety of revealed preference approaches. Those most developed for valuing non-marketed goods probably are the hedonic labor market and property value approaches and the travel cost approach (TCM) to valuing recreation. None of these will be particularly useful for valuing the reproductive effects of concern here. Another approach is the averting behavior approach. This involves placing values on steps individuals take to avoid some "bad" outcome as a proxy (but generally an overestimate of) the WTP to avoid that outcome. For instance, if someone stays indoors with the air conditioner on all day *because* of the high pollution, the added costs of their electricity bill may have some relationship to their WTP to avoid the health effect. For this approach to yield defensible estimates of value requires a number of very stringent assumptions and, therefore, is not often used. However, as seen below, this approach holds some promise for estimating the value of avoiding low birth weight babies.

For valuing goods traded in markets, there are quite well-established valuation techniques. For instance, if reduced pollution raises crop yields, this is modeled as an outward shift in the supply of the crop, meaning that more of this crop can be produced at any given market price. Given the demand for the crop and the assumption that the pollution change affects a substantial portion of the crop, this shift results in a lower price for the crop. As a result, consumers benefit. Producers also benefit because the inputs they use appear more productive. Once the analyst can estimate the shift in supply (from concentration-response functions supplied by, in this case, plant scientists), these effects are easy enough to estimate from data collected on the market price and quantities

⁷ This example forms the basis for the criticism that CBA neglects important distributional considerations.

produced and sold, plus analyses in the literature on the responsiveness of supply and demand to price (price elasticities). Fertility services, in particular, are goods traded in markets. Thus, the social benefit of reducing demand for these services can, in theory, be measured by standard techniques.

There are several additional approaches to valuing health effects. One -- the cost of illness approach -- attempts to tally the various out-of-pocket costs associated with illness. By missing "pain and suffering," this approach necessarily underestimates costs (benefits). Hospitalization, emergency room, doctor and drug costs (including charges paid by insurance companies), the value of non-work time spent in these activities and being sick, and the value of work lost as a result of illness are the categories of costs usually estimated.

A second shortcoming of the cost of illness approach is related to the fact that costs are measured *ex post*, given the existence of a medical condition, rather than *ex ante*. Due to the uncertain nature of future health states, *ex post* cost estimates will likely differ from *ex ante* estimates. Because *ex ante* estimates are the proper basis for analysis, the cost of illness approach does not provide cost estimates that can be directly incorporated into analysis.

While properly performed revealed or stated preference analyses are generally acknowledged to produce valid results, both groups of techniques are subject to limitations. The "constructed choice" nature of stated preference techniques increases the possibility of responses that do not reflect true consumer preferences. Respondents might provide inaccurate information due to poorly understood questions or poorly designed questions that impart strategic behavior on the part of those surveyed. Revealed preference techniques, while based on actual consumer behavior, are restricted to the observed market conditions. As a result, they may be of limited value in situations where the conditions to be analyzed differ substantially from current markets.

An analysis based on a combination of revealed and stated preference data can draw from the strengths of each of the two methods.⁸ Stated preference responses can be used to supplement the revealed preference information in order to examine scenarios that may be removed from current market conditions or to fill in gaps not revealed through market behavior (such as the value of avoiding the anxiety and discomfort associated with fertility treatments). The inclusion of revealed preference information allows for the use of data based on consumer behavior rather than stated responses to constructed choices. Revealed and stated preference data obtained from the same sample have the added benefit of possessing the same underlying preference structure.

To see how these approaches can be used in the context of concern here, consider the options for valuing changes in fertility. One could use *stated preference* approaches,

⁸ See Cameron (1992), Adamowicz, Louviere and Williams (1994) and Eom and Smith (1994) for explanations of the combined revealed and stated preference technique.

where, for example, child-bearing couples suffering from infertility are given the opportunity to pay for cures.⁹ Or, one can rely on *revealed preference* approaches, where infertile couples are observed actually paying for infertility treatments.¹⁰ The dollar cost of these treatments to the couples can then be viewed as a lower bound on the value they place on successful child-bearing.

Using the revealed preference approach, the cost of infertility treatment provides a lower bound on the value of child-bearing for two reasons. First, the cost of infertility treatment (the price) is a measure of value only at the margin and for most couples there will be some consumer surplus. To capture the consumer surplus, one requires a complete demand curve for infertility treatment and this requires one to specify the *choke price*, that is, the price at which no treatment is demanded. In addition, there may be large nonpecuniary costs of undergoing infertility treatment (e.g., the time involved, the reactions to drugs used, etc.).

Finally, a combined revealed and stated preference framework for fertility valuation may be used. Revealed preference information is based on observed market transactions for infertility treatments. However, we can expand the framework to include behavior related to the time and other nonpecuniary cost of treatment not directly observable in market transactions. By broadening the framework to include behavior not recorded in markets, we can generate a more accurate measure of the true cost of infertility treatment. Finally, we can employ stated preference techniques designed to estimate choke prices (Cameron, 1992) in order to capture the full range of welfare effects to those seeking or engaged in treatments. Note, however, that this approach would not capture the effect of increased fertility on those who would not seek treatment. Doubtlessly, some would be willing-to-pay for reducing their expected time for becoming pregnant from, say, six months to five. This approach ignores this group and specifies a more narrow model where benefits of improved fertility only accrue to those who would otherwise be judged clinically infertile.

2.C. Non-Welfare Based Measures of Health Effects

The preceding section has focused on methods of estimating monetary measures of economic value. Such estimates may be used in cost-benefit analysis, where monetary estimates of benefits and costs associated with a policy are compared. An alternate approach toward policy evaluation does not attempt to express both costs and benefits in monetary terms, but instead, compares choices in terms of the cost per effect, such as dollars per life-year gained. As Johannesson and Jönsson (1991) and Johannesson (1995)

⁹ Stated preference approaches are most often associated with the technique of contingent valuation (see Mitchell and Carson, 1989).

¹⁰ Revealed preference approaches rely on the observation of actual past behavior. Thus examining market transactions is a form of revealed preference valuation. In the context of infertility, examining the cost of infertility treatments paid by infertile couples is a revealed preference technique applied to valuation of childbearing.

note, cost-effectiveness analysis has been used to a greater degree than cost-benefit analysis in studies related to health issues.

Since cost-benefit analysis and cost-effectiveness analysis differ in terms of the method in which outcomes are expressed, the two forms of analysis convey different information. Expressing both costs and benefits in common units during the process of cost-benefit analysis allows one to determine whether a policy would result in positive or negative changes in welfare. Cost-effectiveness analysis can rank projects according to effectiveness, but it cannot determine if a project will result in a positive net benefit. Implicitly, one must determine the minimum effectiveness level that is acceptable for project implementation.

Analysis based on the cost per life-year gained does not account for the fact that illness and medical treatments affect not only the quantity but also the quality of life. That is, two treatments may result in a similar quantity of years of life added, but these years may differ in terms of characteristics such as physical functioning, role functioning and mental health. For example, a lower value is associated with a year confined to a bed than with a year of perfect health. For this reason, cost-effectiveness analysis has been refined to account for qualitative differences across health states. Such analysis is sometimes referred to as cost-utility analysis (Johannesson and Jönsson, 1991; Johannesson, 1995)

A prominent method of accounting for differences in the quality of life involves the use of quality-adjusted life-years (QALYs).¹¹ Life-years are adjusted by a quality weight, where full health is usually assigned a value of 1 and death a value of 0. For example, a year of a condition requiring mechanical equipment to walk might be assigned a value of 0.79 while blindness might have a value of 0.39. A medical treatment extending life by 6 years but resulting in blindness during those years would thus be equivalent to 2.34 QALYs.

The theory underlying QALYs does not specify how the quality weights are determined. Weights may not be based on welfare theory and may reflect the preferences of physicians, planners or government officials rather than those who consume the medical services (Torrance and Feeny, 1989; Gafni and Birch, 1995). Even if weights are derived from consumer preferences, QALYs are consistent with welfare theory only if a specific set of conditions are satisfied (Pliskin, Shepard and Weinstein, 1980; Torrance and Feeny, 1989; Wakker, 1996). These conditions include: (i) utility independence between quality and quantity, (ii) constant proportional trade-off between quantity and quality, (iii) risk neutrality with respect to time and (iv) strong separability between qualities at different points in time. These restrictions are not normatively appealing (Gafni and Birch, 1995) and have not been empirically validated (Loomes and McKenzie, 1989).¹²

¹¹ For an overview of QALYs and their relation to utility measures, see Torrance and Feeny (1989).

¹² A recent evaluation of the validity of QALYs is contained in Bleichrodt and Johannesson (1996).

A range of methods, rather than one standard technique, have been used to estimate QALY weights. Common methods include the standard gamble, time trade-off, and rating scales. However, only the standard gamble method has a foundation in von Neumann-Morgenstern expected utility (Torrance and Feeny, 1989). Different methods used to determine weights can result in different estimates of QALYs, resulting in different estimates of cost-effectiveness. For example, Hornberger, Redelmeier and Petersen (1992) found that the cost per QALY saved through in-center hemodialysis varied from \$34,000 to \$45,000 depending on the method used to estimate QALYs.

Healthy-years equivalents (HYEs) has been proposed as an alternate method of accounting for quality differences across health outcomes (Mehrez and Gafni, 1989; Gafni, Birch and Mehrez, 1993; Gafni and Birch, 1995). A two-stage procedure using the standard gamble technique is used to determine the years of full health that are perceived as being equivalent to a given number of years of less than perfect health.

There is currently much debate concerning the value of HYE relative to QALYs. Some studies have professed that HYE are theoretically more attractive than QALYs (Mehrez and Gafni, 1989; Gafni, Birch and Mehrez, 1993; Gafni and Birch, 1995) while others have dismissed HYE as a more complicated method of obtaining the same information included in QALYs (Johannesson, 1995; Loomes, 1995; Wakker, 1996; Weinstein and Pliskin, 1996).

Within the health care field, there has been little research comparing the above nonpecuniary methods of representing preferences to welfare-based measures. In addition, it has not been firmly established which nonpecuniary technique best represents preferences. Examination of these issues should be a focus of future research.

3. Overview of the Remainder of the Report

The following chapters review existing studies that are relevant to an estimation of the economic values associated with the reproductive and developmental processes, describe data sources that may be used in future research, and provide suggestions for future research methods. The remainder of the report begins in chapter 2 with an examination of existing analyses. As described above, one possible approach toward deriving value estimates involves the use of stated preference techniques. Several studies have attempted to determine couples' willingness to pay for infertility treatments through contingent valuation surveys, and these studies are reviewed below. The use of revealed preference information to determine value estimates, such as the responsiveness of infertility treatment utilization to changes in treatment cost and use of adoption services, is also discussed. Finally, estimates of the cost of a specific infertility treatment, in vitro fertilization, are described. Existing studies related to low birth weight and birth defects are limited to cost of illness analyses. Although cost of illness analyses only provide a lower bound estimate of the true cost of these endpoints, estimates based on techniques

that can provide more accurate welfare measures, such as contingent valuation surveys, do not exist.

Chapter 3 focuses on data sources containing information that may be used in future analysis. In addition to the cost of infertility treatment information described in chapter 2, data exist regarding the prevalence of infertility and utilization of infertility treatments. While infertility prevalence information is largely derived from a single series of surveys, the National Survey of Family Growth, data regarding the prevalence of low birth weight or birth defects can be obtained directly from vital statistics records. Other relevant low birth weight and birth defects data sources include surveys and monitoring programs which contain information related to actions taken by parents, such as smoking, that affect the likelihood of occurrence of these endpoints. Such information can be used to derive estimates of the value placed on a healthy birth.

Recommendations for future research are provided in chapter 4. The infertility treatment process is discussed and a preliminary model describing a couple's participation in the process is developed. Aggregation of couples' decisions results in demand curves for infertility treatments. Since estimation of infertility treatment demand curves involves the use of both market data and unobserved preference information, a combined revealed and stated preference approach is recommended. Welfare estimates can then be derived from these demand curves.

Recommendations for future research regarding low birth weight and birth defects focus on specifying the proper concept of cost associated with the occurrence of these conditions. Given factors that differentiate these conditions from existing analyses of morbidity, development of a formal economic model is recommended. Estimation of the true costs of low birth weight and birth defects may be obtained through techniques such as contingent valuation or an examination of actions taken by parents that affect the likelihood of a healthy birth.

Chapter 2 Literature Review

1. Introduction

The economic and medical literature was reviewed to assemble the existing research providing estimates of the value of reducing the incidence of human reproductive and developmental abnormalities. The results of this review are presented below by endpoint. Within each endpoint, articles are sorted according to the method used to obtain a value estimate.

Our literature review considered the following end-points; infertility, low birth weight and birth defects. Within each end-point category we reviewed studies that utilize one of three approaches to economic valuation: stated preference, revealed preference and cost-of-illness.

2. Infertility

Infertility is defined as the inability to conceive after 12 months of intercourse without contraception. Using this definition, the rate of infertility for U.S. couples between the ages of 15 and 44 was about 7.9 percent or one couple in twelve in 1988.¹³

There is a very small body of existing research related to the value of infertility treatment services. A limited amount of studies have provided either direct estimates or information that can be incorporated into further research. Most of the directly applicable analyses are in the form of stated preference surveys. Additional existing studies, while not actually providing value estimates, can aid in an evaluation of revealed preference or cost of illness techniques.

2.A. Stated Preference

2.A.1. Review of Existing Studies

There are a handful of studies that have used stated preference techniques to estimate the value of infertility treatments. The majority of these have expressed the outcomes in terms of willingness to pay (Neumann and Johannesson, 1994; Ryan, 1994; Granberg et al., 1995), while one study has expressed preferences in nonmonetary terms (Goeree, Labelle and Jarrell, 1993b). The primary goal of the willingness to pay studies was to introduce a new technique that might aid in the evaluation of a rapidly developing infertility treatment rather than to provide an exact estimate of willingness to pay through rigorous analysis. As a result, details of survey techniques that would aid in an evaluation of the studies are not always reported. Details that are reported or that can be inferred indicate that there are significant shortcomings with existing stated preference surveys

¹³ See Mosher, W. D. and W. F. Pratt. 1990. "Fecundity and Infertility in the United States, 1965-88," *Advance Data*, Number 192, December 4, National Center for Health Statistics, U.S. Department of Health and Human Services, Public Health Service.

related to infertility treatment. The scarcity of existing research and the limited applicability of current work highlight the need for additional research in this area.

The size or composition of the samples used in the existing surveys precludes the application of many of the results to broader research. Samples used in several of the studies, had demographic characteristics that were not representative of the general population (Neumann and Johannesson, 1994; Granberg et al., 1995). As a result, preferences for infertility treatment as indicated by the willingness to pay for these respondents may not reflect the value placed on treatment by the general population. While Neumann and Johannesson (1994) and Ryan (1994) utilized samples of over 100 respondents, the small sample size used in Granberg et al. (1995) may limit the statistical reliability of the results of that study.

If performed properly, the contingent value technique is generally accepted as a method of determining preferences toward a good. Care must be taken to ensure that the survey questions result in responses that truly reflect preferences. Due to the constructed choice nature of the CV surveys, care is needed to ensure that results are reflective of respondent preferences. The CV survey formats used in the studies valuing infertility treatment we have reviewed are, in our opinion, not in general agreement with CV methods and protocols designed to yield reliable measures of economic value. For example, Granberg et al. (1995) used a open-ended CV value elicitation format rather than the more widely accepted dichotomous choice format. Neumann and Johannesson (1994) and Ryan (1994) both note that the combined open-ended and closed-ended question methodology that is used in the two surveys might be subject to anchoring bias, a condition where the response is influenced by the alternatives offered. Anchoring bias was tested for and found to exist in some questions by Neumann and Johannesson (1994). As the authors note, a survey format where participants accept or reject a single offer is less susceptible to this problem.

In addition to the impact that the structure of the questions may have had on responses, respondents' answers may have also been influenced by the existence of insurance coverage for treatment. The average willingness to pay estimated by Ryan (1994), \$2,506 Australia dollars, was very close to the estimated government expenditures on Medicare and average drug use, \$2,157 Australia dollars. In the study performed by Granberg et al. (1995), 40 percent of the respondents came from public clinics. The fact that as the price of an episode of treatment was hypothetically altered respondents altered the quantity demanded so as to keep overall expenditures fairly constant, may partially reflect the effect of treatment coverage. If respondents were aware of the limits of treatment coverage, willingness to pay responses may have been influenced by degree that costs were covered by third parties. Since both Ryan (1994) and Granberg et al. (1995) surveyed individuals who were currently undergoing or had undergone infertility treatment, it is likely that participants had a knowledge of the limits of treatment coverage.

While there have been several preliminary surveys of willingness to pay for in vitro fertilization (IVF), no existing analysis has used stated preference techniques to examine

the willingness to pay for other more common infertility treatments. To the extent that the treatment method itself impacts valuation, a more comprehensive analysis of different treatments may be necessary for a complete estimate of the value of the infertility treatment process.

2.A.2. Summary of Existing Studies

Neumann and Johannesson (1994) developed a CV survey in which respondents were asked several different hypothetical questions related to IVF treatment. In one scenario, respondents were asked how much they would be willing to pay for an IVF cycle given the fact that they were infertile (defined as the ex post value of IVF). The respondents were told that adoption was available at a cost of \$15,000.

A second scenario framed the question of willingness to pay in terms of medical insurance. Previous studies have noted that more reliable results may be obtained when medical valuation questions are framed in terms of insurance payments since this more closely reflects the type of cost that is incurred for treatment (Gafni, 1991). Respondents were asked how much they would be willing to pay, not knowing their fertility status, for a one time option to purchase insurance that would provide IVF services if needed (defined as the ex ante value of IVF). They were told that 10% of couples are infertile. Private IVF services would cost \$25,000 if insurance was not purchased and the services were needed.

Third, all respondents were asked how much they would pay through higher taxes for a public IVF insurance program. This attempted to capture an altruistic element in the valuation of IVF, since not all who paid taxes would benefit from the IVF program.

Finally, respondents were asked to compare infertility risk reduction with mortality risk reduction by choosing between two uses of public funds: providing IVF coverage for state residents or reducing highway fatalities. As the number of deaths avoided was varied from 1 to 500, respondents were asked which program they would choose given that the IVF program would result in 300 births.

On average, respondents were willing to pay \$17,730 for IVF treatment having a 10% chance of success. Ex ante, individuals were willing to make a one time payment of \$865 for insurance providing IVF if needed. The study also found that individuals would be willing to pay \$32 per year in taxes for a public program giving 1,200 couples per year a 10% chance of successful fertilization. A program resulting in 300 IVF babies was equivalent to one reducing auto deaths by 35 per year.

The average response values for each of the scenarios were combined with the corresponding probabilities in order to derive estimates of the willingness to pay per statistical baby. These willingness to pay estimates were determined to be \$177,300

ex post, \$1,730,000 ex ante and \$896,000 for the public insurance program funded through higher taxes.¹⁴

Ryan (1994) surveyed couples in order to gain an understanding of the psychological elements associated with infertility treatments. A willingness to pay question was included in the survey in order to capture individuals' overall valuation of treatment.

A questionnaire was sent to 700 women and 200 male partners enrolled at a private infertility treatment service in Sydney, Australia. Respondents were asked to rate various attributes of the assisted reproductive technology (ART) program, such as information, counseling and follow-up support, on a scale of 0-10. Respondents were also asked to rate a series of statements intended to capture feelings of regret and disappointment towards the infertility treatment.

Participants who were undergoing ART treatment were asked to state their maximum willingness to pay for the current treatment. Participants who were no longer undergoing treatment and who were not willing to undergo another attempt were asked to state their maximum willingness to pay for the last attempt at IVF. This was viewed as providing willingness to pay measures under certainty and uncertainty. Willingness to pay measures for those undergoing treatment included those who had just begun treatment as well as those who had undergone several attempts. The mean and median willingness to pay for an episode of treatment was AU\$2,506 and AU\$2,250, respectively.

As part of a cost-benefit analysis of IVF, Granberg et al. (1995) estimated willingness to pay for couples at two Swedish clinics from January 1992 to March 1993. Forty-seven couples were asked to state the number of cycles they would be willing to undergo as the price of treatment was hypothetically altered. In addition, respondents were asked to state their maximum willingness to pay for having a child. Fifty-five percent of the couples were willing to pay at least £10,000 (about \$17,600) to have a child.

Goeree, Labelle and Jarrell (1993b) used several methods other than willingness to pay to gain insight into individuals' valuation of IVF. They surveyed 80 couples enrolled in an IVF treatment program study and 80 members of the local population in Hamilton, Ontario.

Respondents were asked to rank a series of 9 health outcomes with choices such as "no procedure, spontaneous pregnancy, healthy child", "successful procedure, twin

¹⁴ Ex post, an episode of treatment was assumed to have a 10 percent chance of success. Thus, on average 10 episodes of treatment would result in one successful outcome. Ex ante, 10 percent of couples were assumed to be infertile. 5 percent of these couples were assumed to be eligible candidates for IVF treatment having a 10 percent chance of success. The corresponding ex ante probability is thus $.0005 = .05 \times .1 \times .1$. For the public insurance program, this probability was converted to an annual probability by assuming that on average there were 14 years during which respondents were at risk of using IVF. The corresponding probability is thus $.000036 = .0005 \div 14$.

pregnancy, both healthy" or "no procedure, no pregnancy, remain childless" on a scale of 0 (death) to 100 (perfect health). Respondents in the IVF treatment were surveyed initially and after a six month interval.

In general, respondents in the community sample rated the IVF health outcomes significantly higher than the experimental or control groups in the IVF study. Much of the higher ranking could be explained by the fact that the community sample ranked non-child outcomes, such as "unsuccessful procedure, no pregnancy, remain childless," higher than the IVF study groups.

Respondents' valuations of the various outcomes were used with probability estimates of the possible outcomes to derive expected utility results for "IVF treatment" and "no IVF treatment." Expected utility was calculated separately based on community sample valuations and IVF study groups' valuations.

Sensitivity analysis indicated that there was no significant difference in expected utility between "IVF treatment" and "no IVF treatment" when either the community sample's or the IVF patients' valuations were used.

Valuations of the possible health outcomes were compared over time in order to test the hypothesis that the IVF procedure has a value regardless of the outcome because it allows patients to feel that they have done all that is possible and "get on with their lives." Comparison of the experimental IVF study group's health outcome rankings during the initial interview and during the six month follow-up did not indicate any change in valuations after the treatment, refuting the "treatment effect" hypothesis. The authors note, however, that the follow-up time period may have been too short to capture this effect.

Community members were asked to rank the value of an IVF program relative to other health care programs such as rape crisis centers, school immunization programs and childhood cancer treatment centers. Since the rankings were ordinal, survey results were presented in the form of the number of times each program was ranked highest or lowest and the number of times it was ranked in the top or bottom three responses.

IVF treatment was consistently ranked below other programs, above only prenatal classes. Twenty-five respondents ranked IVF the least important and fifty ranked IVF among the three least needed programs.

2.B. Revealed Preference

2.B.1. Insurance Coverage of IVF

Medical insurance reduces the cost of treatment incurred by patients. Differences in insurance coverage for IVF result in differences in treatment costs across couples

seeking infertility treatment. An analysis of the fraction of the cost paid by couples with varying degrees of insurance coverage and the corresponding utilization of IVF services could aid in estimating the demand for these services.

Collins et al. (1995) used aggregate statistics to derive a rough estimate of the relation between the utilization of IVF services and the level of patient copayments. It was estimated that country-wide average copayment levels were 85%, 15% and 7% in the U.S., Ontario, Canada and France, respectively. Using this information and information concerning IVF cycles performed per million population, estimates of elasticity of utilization with respect to copayment were found to be between 2.2 and 4.3.

The authors note that the estimate is much larger than previous health care utilization elasticity estimates. A true measure of elasticity should relate utilization of IVF services by those enrolled in the insurance program and the copayments levels of the program. Country-wide aggregate statistics concerning cycles performed do not identify treatment recipients. Treatment received by couples not covered by the insurance program would cause the utilization estimate to be overstated.

There has been no other study which analyzed the relation between copayment levels and utilization, although general statements indicate that utilization increases with decreases in copayment levels. Bartels (1993) noted that in Australia, where the government paid for 70% of the IVF treatment cost, 519 treatments per million inhabitants occurred in 1987. In Britain, where the government paid a smaller fraction of the cost, 117 treatments per million inhabitants occurred in 1986.

Within the U.S., several states have passed laws requiring insurance companies to offer or provide coverage for infertility treatment. This legislation might result in interstate variation in treatment utilization. Ideally, one could compare utilization of services in states where infertility treatment coverage is mandated with utilization of services in states having lower levels of insurance coverage. Differences in utilization resulting from differences in the fraction of the cost paid by couples could indicate how much infertile couples are willing to spend on the opportunity to have a child.

There are several factors complicating an interstate insurance analysis. Aggregation of the analysis to the state level may not reveal a significant relation between copayment level and utilization since the laws do not significantly and uniformly lower the cost of treatment relative to states without mandated coverage. For example, all couples who potentially might make use of infertility treatment services are not covered in states mandating coverage. Pratt et al. (1994) indicated that the Family Building Act in Illinois excludes between 25% and 40% of couples who are in their reproductive years.

A more serious difficulty arises from the fact that one cannot generally assume that a correlation between favorable insurance coverage and utilization of IVF services implies a causality running from level of insurance coverage to utilization. Couples planning to use infertility treatment services are more likely to select insurance programs offering

better coverage and there is thus a nonrandom distribution of couples across insurance plans. This problem of adverse selection stems from the fact that couples have more information concerning their medical needs than insurance companies.

Leibowitz (1990) analyzed the effect of changes in insurance coverage on the decision to have children. The adverse selection problem was avoided by the use of data from the RAND Health Insurance Experiment (HIE) which randomly assigned individuals to different insurance plans and provided various forms of coverage for three to five years. As a result of the random assignments, the findings of the experiment were free of any self selection bias.

Medical coverage was assumed to influence the child-bearing decision since the cost of medical care has been estimated to be five to nine percent of the total discounted cost of rearing a child (Lindert, 1978). Women who were assigned to free medical care had 29 percent more births during the experiment than women who were assigned to plans requiring cost sharing. The birth rate of women in the free plan (76.9 per 1,000) was above the national average at the time (65 to 68.4 per 1,000) while the birth rate of women assigned to the coinsurance plan was below the national average. This was seen as evidence of the fact that a temporary change in the cost of medical care caused participants to alter the timing of the child-bearing process. Women who were given a temporary discount in the cost of medical care through enrollment in the free plan were more likely to have children while women assigned to the coinsurance plan were more likely to delay child-bearing.

It appears that adverse selection is a difficult problem to overcome in analyzing utilization of services covered by private insurance. Collins et al. (1995) included IVF utilization rates in Ontario, Canada due to favorable IVF coverage that existed there in 1993. Collins (1994) noted that in April 1994 coverage of IVF in Ontario was limited to women with bilateral tubal obstruction. If sufficient data existed, one could compare utilization rates before and after the legislation limiting coverage. This analysis might be insightful if the presence of government-funded health care lessened the problem of adverse selection.

2.B.2. Infertility Treatment and Adoption

Examination of data related to adoption services used by couples who had previously attempted infertility treatments might aid in estimating the demand for children. Unfortunately, there is very little demographic data related to adoption services. National estimates of all types of adoption have not been collected by the government since the mid-1970s. Like infertility, adoption is not common among the general population and surveys require large samples in order to obtain meaningful statistics.

2.C. Cost of Illness

Infertility treatment cost information can be used in several different methods of value estimation. Direct medical costs of treatment can be combined with estimates of the cost of time spent at treatment in order to form a cost of illness estimate, which is a lower bound estimate of the true value of treatment. Alternatively, information concerning the actual price of treatment facing couples and the resulting utilization of treatment can be supplemented with stated preference information describing treatment demand at other prices in order to estimate demand curves for infertility treatments and the corresponding welfare measures.

Table 3.1 summarizes the results of studies that have estimated the cost of IVF. Differences in cost estimates exist because there is not one standard formula that is used to measure the cost of undergoing IVF treatment. Rather, there are a range of factors that account for differences among the estimates. For example, estimates vary according to the unit on which costs are based. Cost estimates may be based on a single episode of IVF treatment, referred to as a cycle. A complete cycle involves use of fertility drugs to stimulate egg development, retrieval of eggs from the female's ovaries, fertilization of the eggs in a laboratory and implantation of the fertilized eggs into the female's uterus (Neumann et al., 1994). Differences exist even among analyses based on the cost per cycle, since not every cycle that is initiated is completed. Each stage of the cycle is associated with a chance of failure and costs are lower for incomplete cycles, as the entire medical procedure is not performed. The cost per initiated cycle thus will usually differ from the cost per completed cycle.

Alternatively, since a healthy child is the desired outcome of treatment, costs may be expressed in terms of cost per birth. If an IVF cycle is estimated to have a ten percent chance of success, ten cycles on average will result in one successful outcome. Analyses based on the cost per statistical birth are subject to more uncertainty than those based on the cost per cycle because the former cost requires an estimate of the success rate for an individual cycle of treatment. Success rate estimation is complicated by factors such as treatment-independent pregnancies and substantial treatment drop out rates after each failed cycle. Success rates also vary according to the underlying cause of infertility. They are highest for tubal infertility and lowest for infertility caused by male factors (Schenker, 1993). This implies that the cost per statistical birth would vary according to the way in which pollution affects the reproductive process.

In some circumstances, the social cost of providing a good or service may not equal the private cost of purchasing the good or service. This is especially true in the area of health care where charges may not equal costs due to medical insurance. Costs estimates for IVF may be based on charges or average discount rates rather than actual economic costs due to the fact that treatment of IVF is partially covered by health insurance. When using cost estimates from existing studies, it is important to determine whether the study has estimated the social cost or the charge facing a couple undergoing treatment. Cost of illness analysis should incorporate the total costs incurred by society.

Table 3.1 - Cost of IVF

Author	Cost	Source
Neumann, Gharib and Weinstein (1994)	\$8,000 per IVF cycle \$50,000 to \$800,000 per delivery	charges contained in published brochures from six in vitro fertilization centers in the U.S.
Collins et al. (1995)	\$7,861 per completed IVF cycle	survey of clinics listed in Society for Assisted Reproductive Technology database
Goldfarb et al. (1996)	\$39,249 per delivery for single and twin pregnancies \$342,788 per delivery for triple and quadruplet pregnancies.	patient records maintained during 1991 and 1992 at the University Hospitals of Cleveland IVF program
Goeree, Labelle and Jarrell (1993a)	CA\$3,937-CA\$5,950 average incremental cost of IVF versus other treatment or no treatment	Chedoke-McMaster's fertility clinic in Hamilton, Ontario
Stern et al. (1995)	\$19,267 per IVF baby	expenditures from Hadassah University Hospital in Jerusalem
Page (1989)	\$17,000 per maternity	two Regional Health Authorities funding IVF services and a private IVF unit in the UK
Granberg et al. (1995)	\$6,900 per started treatment \$20,300 per delivery	one private and one public IVF clinic in Sweden
Haan (1991)	\$1,000 to \$1,650 average cost per treatment	one general and four university hospitals covering IVF treatments in the Netherlands

Estimation of treatment demand curves should account for the fact that couples' decisions are based on costs net of insurance reimbursement.

In addition to the effect that insurance has in altering estimates of the cost of IVF treatment, estimates may differ due to what is incorporated in the definition of "cost." Some estimates, for example, have included the opportunity cost of the couple's time while others have not.

A final source of differences among the cost estimates is due to the fact that the IVF treatment process has been characterized by substantial changes in technology over time, which has led to changes in procedures used and the costs of the procedures. While most of the cost estimates presented in table 3.1 were based on costs occurring at similar points in time, differences in cost estimates occurring at different times can be partially explained by changes in technology.

A portion of the IVF cost estimates listed in table 3.1 are based on the IVF treatment process as it occurs in countries other than the U.S. (Page, 1989; Haan, 1991; Goeree, Labelle, Jarrell, 1993; Granberg et al., 1995; Stern et al., 1995). Differences in health care systems and differences in procedures and technology across countries have led to a wide range in international IVF cost estimates (Stern et al., 1995). These differences may limit the applicability of foreign estimates of the cost of IVF in an estimation of the cost of IVF treatment facing couples in the U.S.

In summary, cost estimates for IVF treatment differ due to differences in bases, treatment of insurance, definition of cost, and technology. Differences in treatment coverage, technology and procedures across countries limit direct international comparisons.

It is important to note that the majority of infertility treatment cost analyses have focused on the IVF technique. The recent examination of IVF has occurred in response to the need to determine the cost-effectiveness of an expensive technique that has grown in popularity despite uncertain success rates. According to the American Society for Reproductive Medicine (ASRM), IVF is used to treat women with fallopian tube damage, endometriosis, male factor infertility, and couples suffering from unexplained infertility who have not been successfully treated with other procedures. Despite this range of applications, most couples undergoing infertility treatment are not treated with IVF. Wilcox and Mosher (1993), using data from cycle IV of the National Survey of Family Growth, indicated that only 2% of women with reproductive difficulties reported having undergone IVF treatment. Drugs used to induce ovulation were a much more common treatment, used by 20% of respondents. The fact that the majority of existing cost information is related to a technique used in a minority of treatment cases highlights the need for an analysis of more common procedures.¹⁵

¹⁵ Cooper (1986) provided an early estimate of the cost of infertility treatments where costs per pregnancy were estimated for specific diagnoses. The average costs per pregnancy for ovulation defects, seminal

3. Low Birth Weight

Low birth weight (LBW) is defined as 2500 g or less, very low birth weight (VLBW) as 1500 g or less and extremely low birth weight (ELBW) as 1000 g or less. LBW is a major cause of neonatal and infant mortality in the U.S. Low birth weight survivors are more likely to have health problems than those born at a heavier birth weight. In addition, they are more likely to experience preschool developmental delays and additional adverse effects later in life (Chaikind and Corman, 1991).

The development of neonatal intensive care has reduced the neonatal mortality rate over time, especially among newborns having the lowest birth weight. In 1960 in the U.S., 10,241 white singleton babies weighing less than 1000 g were born and 67 survived. In 1983, 3,840 of 8,542 such infants survived (Paneth, 1995). Despite improvements in the ability to keep low birth weight infants alive, the rate of infants born with low birth weight has not declined over time.

Low birth weight occurs disproportionately with poor, poorly educated and black mothers. Shiono and Behrman (1995) note that only seventeen percent of all births are to African-American families, but these families account for thirty-three percent of all LBW and thirty-eight percent of all VLBW births. As a result of differences in incidence among racial and socioeconomic categories, an analysis of the factors influencing the incidence of LBW should control for these variables.

3.A. Stated Preference

No studies have been found which use stated preference techniques to value the benefits of avoiding LBW.

3.B. Revealed Preference

No studies have been found which use revealed preference to estimate the benefits of avoiding LBW.

3.C. Cost of Illness

Schwartz (1989) estimated the likelihood of occurrence of different birth weights and the immediate in-hospital costs associated with each weight category. Data was collected from 28 perinatal centers that provided tertiary care. Tertiary care was defined as having a neonatal intensive care unit, retaining all neonates who required treatment, and

factors, and tubal factors were estimated to be \$5,041, \$8,811 and \$31,841, respectively. These cost estimates, however, are of limited value due to the significant changes in technology that have occurred since the early 1980s. Goeree, Labelle and Jarrell (1993a) also estimated the costs of several non-IVF treatments, but these estimates are based on the Canadian health care system.

having at least one full-time neonatologist on staff. The sample was found to be representative of the 360 urban tertiary hospitals in the U.S.

Financial data based on charges (the amount billed each patient for the hospital services) was provided by the participating hospitals. Charges were converted to costs and expressed in terms of 1985 dollars.

Table 3.2 - Survival Rates, by Birth Weight

Birth Weight	Total Quantity of Births	Quantity Survived	Percent Survived	Quantity Died	Percent Died
all	80,282	77,491	97	821	1
500-749g	384	90	23	273	71
750-999g	531	274	52	163	31
1,000-1,249g	577	351	60	84	15
1,250-1,499g	701	487	69	40	6
1,500-1,999g	2,121	1,636	77	61	3
2,000-2,499g	4,622	4,275	93	49	1
≥2,500g	71,346	70,378	99	151	.2

Source: Schwartz (1989)

Table 3.3 - Cost of Inpatient Hospital Care Among Surviving Neonates

Birth Weight	Percent of Patients	Average Cost Per Patient	Percent of Distribution
all	100	\$1,449	100
≤2,500g	9.2	9,072	57.5
500-749g	.1	64,161	5.1
750-999g	.4	45,336	11.1
1,000-1,249g	.5	28,486	8.9
1,250-1,499g	.6	19,497	8.5
1,500-1,999g	2.1	9,695	14.1
2,000-2,499g	5.5	2,568	9.8
≥2,500g	90.8	678	42.5

Source: Schwartz (1989)

Table 3.2 indicates that newborns having the lightest birth weight are at the highest risk of dying. Table 3.3 illustrates that the cost of hospital care is disproportionately incurred by infants who are LBW. Low birth weight infants represent nine percent of neonates who went home from the hospital but involved fifty-seven percent of the total acute inpatient cost for infants.

Cost savings were estimated for the universe of urban tertiary hospitals, which care for fifty-three percent of low birth weight infants, by assuming that twenty percent of all infants were shifted to the next heavier weight category. Overall savings were estimated to range from \$73-\$96 million for 1985. Infants weighing less than 1500g, 1.6% of all infants, accounted for 42% of the savings.

The analysis by Schwartz (1989) indicates that there is a nonlinear relation between birth weight and cost, with the smallest infants accounting for the majority of the cost. Increases in VLBW or ELBW infants would have a greater impact on costs than an increase in heavier low birth weight infants. As a result, in analyzing the increased incidence of low birth weight due to pollution, it is important to specify whether there is a proportional increase among all low birth weight categories or whether some categories increase more than others.

Table 3.4 - Incremental Direct Costs of Low Birth Weight Among Children from Birth to Age 15 in 1988

Age Group	Cost Type	Mean Cost per Low Birth Weight Child	Number of Low Birth Weight Children	Total Cost
Infancy	Health Care	\$15,000	271,000	\$4,000,000,000
1 to 2 years	All	not estimated	500,000	not estimated
3 to 5 years	Health Care	290	820,000	240,000,000
3 to 5 years	Child Care	180	820,000	150,000,000
6 to 10 years	Health Care	470	1,300,000	610,000,000
6 to 15 years	Special Education	150	2,400,000	360,000,000
11 to 15 years	Grade Repetition	45	1,100,000	50,000,000
Total			4,000,000	5,400,000,000

Source: Lewit et al. (1995)

Lewit et al. (1995) provided a comprehensive estimation of the cost of LBW for children aged 0-15. The authors found that in 1988 the incremental health care, education and child care cost of the 3.5 to 4 million children aged 0 to 15 born low birth weight (about 7% of all children in that age group) was between \$5.5 and \$6 billion.

Cost estimates were divided into the first year of life, preschool years, and school aged years. Costs included health care costs for infants, health care and child care costs for preschool children 3 to 5 years old, and health care, special education and grade repetition costs for school aged children 6 to 15 years old. Health care costs were based on the 1987 National Medical Expenditure Survey, a national survey of 35,000 individuals in 4,000 households in the U.S. Health care utilization by infants was derived from CIGNA Corporation's national survey. The remainder of the analysis was largely derived from the 1988 Child Health Supplement of the National Health Interview Survey (CHS-NHIS) and the 1991 National Household Education Survey's (NHES) Pre-primary and Primary Surveys. The CHS-NHIS was used for estimates of the use of health care and special education services and grade repetition while the NHES was used for estimates of the use of preschool programs and maternal labor force participation.

No costs were estimated for LBW survivors aged 1 to 2 or for the cost of mortality attributed to LBW. In addition, no estimates were made for the morbidity costs of LBW beyond age 15. As an indication of the costs of morbidity beyond age 15, the authors cite previous research which indicated that if the number of disabled LBW infants in the U.S. were reduced by one half, the present value of the additional lifetime wages would be between \$.9 and \$1.9 billion in 1985 (Chu, 1988).

The estimation of resource utilization attributable to birth weight category accounted for demographic factors such as income and mother's education. Since LBW is more likely to occur in low socioeconomic groups, analysis based on simple correlation would not accurately describe the effect of birth weight on resource utilization.

In 1988, 3.6 million babies were born weighing over 2,500 g, 246,919 weighing 1,000-2,499 g and 23,762 weighing less than 1,000 g. \$4 billion (35%) of \$11.4 billion spent on health care for infants in 1988 was attributed to incremental costs due to LBW. This equaled \$15,000 additional for each of the 271,000 infants born weighing less than 2500 g.

Studies have shown that LBW children often perform poorly in psychological and developmental assessments during their preschool years, but not much is known concerning how this translates to resource utilization. Most studies have looked at narrowly defined cohorts or specific institutions rather than population based measures.

Summarizing results found in earlier work (Corman, 1994), Lewit et al. (1995) note that LBW children aged 3-5 are almost twice as likely to be hospitalized and spend more time in the hospital once admitted than normal birth weight children. As a result, LBW children aged 3-5 spend almost 4 times as much time in the hospital, averaging .23 days per year vs. .06 days per year for normal birth weight children. The incremental cost per child for health care was estimated to be \$290 in 1988, leading to an aggregate incremental cost of \$240 million.

Even though the analysis did not find any effect of LBW on a mother's decision to work or on use of child care services, LBW did have an impact on the quality of preschool instruction. The authors found that LBW children who attend preschool tend to enroll in programs with higher adult-child ratios. If costs are proportional to adult staff-child ratio, LBW would account for an incremental cost in 1988 of \$180 per LBW child aged 3-5, or \$150 million in aggregate.

Studies of school aged children have found that LBW children have average health and function normally in school, but there is evidence of greater neurologic impairments or chronic health conditions and a greater likelihood of experiencing difficulties in school.

A conservative approach was taken in estimating the health care cost of school aged children and costs estimates were limited to increased inpatient hospital care among children ages 6-10. The mean annual number of nights in a hospital for all 6-10 year olds was .12. LBW children in this age group spend .5 more nights per year than NBW children. This lead to an incremental cost per child of \$470, or an aggregate incremental cost of \$610 million.

Schooling cost estimation focused on differences between LBW and NBW children in the use of special education and the amount of grade retention since these are two areas with large cost implications. Previous analysis (Chaikind and Corman, 1991)

indicated that LBW children were almost fifty percent more likely to be enrolled in special education classes than NBW children. This resulted in an estimated incremental cost of \$150 per LBW child or \$360 million in aggregate.

Corman and Chaikind (1993) have found that LBW children are more likely to repeat a grade by grade 10 (31% vs. 26%). Using information concerning the increased likelihood of grade repetition due to LBW and the cost of grade repetition, Lewit et al. (1995) estimated the incremental cost of grade repetition due to LBW to be about \$50 million per cohort. In addition to this direct cost, grade retention may be correlated with additional costs associated with higher drop out rates. No attempt was made to quantify these costs.

The total incremental costs of LBW for children aged 0-15 were estimated to exceed \$5.4 billion in 1988. Almost seventy-five percent of these costs were due to health care costs of infants.

A series of articles examining the causes of neonatal mortality have accounted for the indirect effect of LBW. Joyce et al. (1986) extended the analysis to provide an estimate of the impact of air pollution on race-specific neonatal mortality rates (deaths within the first 27 days of life per thousand live births).

The results were based on an underlying model in which parents' utility was assumed to depend on consumption, the number of births and the survival probability of each offspring. The number of births and the probability of survival were modeled as endogenous variables. The survival probability production function depended on endogenous variables such as the quantity and quality of medical care, maternal cigarette smoking, the use of abortion services and the use of family planning services and exogenous variables such as the reproductive efficiency of the mother and the quality of the environment.

Neonatal mortality rate production functions were estimated using county level data from 677 counties in the white dataset and 357 counties in the black dataset. These counties represented about eighty percent of the black and white population in 1970. Separate equations were estimated according to race because black infant health levels differ significantly from white levels. Air pollution measurements for five pollutants were obtained from EPA's SAROAD. Because data was not available for all five pollutants in all counties, five subsamples were defined according to whether a county had data for one pollutant regardless of the information on the others. A sixth subsample included counties reporting information on all five pollutants.

Results were presented for the neonatal mortality rate production functions for blacks and whites using ordinary least squares and two-stage least squares. The coefficients were generally significant and of the predicted sign. Of the five pollutants examined, sulfur dioxide was found to have the most consistent negative impact on neonatal mortality. Sulfur dioxide was found to affect neonatal mortality directly and

indirectly through LBW. The authors conclude that the regressions show a significant impact of pollutants on neonatal mortality since the analysis controlled for health care factors such as neonatal and prenatal care as well as socioeconomic risk factors (through LBW).

Estimates were obtained for the marginal willingness to pay, defined as the amount of income that must be taken from an individual in order to hold utility constant when the level of pollution declines. The benefits of a 10% reduction in sulfur dioxide ranged from \$54 million to \$1.09 billion (in 1977 dollars).

4. Birth Defects

4.A. Stated Preference

No studies have been found which use stated preference techniques to value the benefits of avoiding birth defects.

4.B. Revealed Preference

No studies have been found which use revealed preference to estimate the benefits of avoiding birth defects.

4.C. Cost of Illness

Table 4.5 - Incremental Cost of Birth Defects in California, 1988, (\$ 000s)

Condition	Medical Cost	Developmental Services	Special Education	Mortality	Morbidity (Unable to Work)	Morbidity (Limited Work)	Total
Spina bifida	22,402	225	5,264	18,615	11,869	-	58,375
Truncus arteriosus	11,784	-	76	11,905	721	195	24,681
Transposition/DORV	18,220	-	556	36,611	5,318	1,499	62,204
Tetralogy of Fallot	20,278	-	502	15,621	4,750	1,277	42,428
Single Ventricle	6,754	-	110	12,573	1,050	284	20,771
Cleft lip/palate	10,639	372	2,217	44,979	16,579	11,568	86,354
TE fistula	6,743	-	-	13,067	-	-	19,810
Atresia, small intestine	6,918	-	-	5,925	-	-	12,843
Colorectal atresia	6,393	-	-	20,470	-	-	26,863
Renal agenesis	2,707	-	-	50,458	-	-	53,165
Urinary obstruction	5,070	-	-	32,276	-	-	37,346
Upper limb reduction	1,220	-	3,067	11,441	5,565	-	21,293
Lower limb reduction	1,814	-	1,497	7,520	9,034	962	20,827
Diaphragmatic hernia	6,876	-	-	38,095	-	-	44,971
Gastroschisis	5,972	-	-	6,852	-	-	12,824
Omphalocele	3,053	-	-	13,154	-	-	16,207
Down syndrome	30,528	12,004	37,132	53,525	76,255	19,286	228,730
Cerebral palsy	93,306	27,525	28,639	26,434	109,182	7,044	292,130

Source: Waitzman et al. (1994)

Waitzman et al. (1994) estimated the incremental cost of 17 of the most clinically significant birth defects and cerebral palsy. An incidence approach was used to estimate lifetime costs stemming from these conditions for individuals born in California in 1988.

Cost estimates were based on direct medical and special service costs and indirect costs of increased mortality and morbidity. Medical costs included inpatient, outpatient and long-term care costs. Special services were comprised of developmental services such as day care centers and counseling and special education. Mortality and morbidity costs were represented by lost productivity as indicated by the Survey of Income and Program Participation and the California Age-Sex Earnings Profiles.

The total cost of a birth defect was defined as the discounted sum of all of the component incremental direct and indirect costs. A given component incremental cost in year j was defined as the product of the per capita cost of resources used by affected individuals of age j and the estimated number of the cohort alive at age j . Incidence, prevalence and cost estimates were obtained from 11 data sources, some of which were specific to California.

As table 5 indicates, assuming a 5% discount rate, overall costs ranged from \$12 million for gastroschisis, a condition with a normal survival rate after the first year and average postinfancy disability rate, to \$292 million for cerebral palsy, which has increased mortality rates throughout life and higher rates of postinfancy disability.

Waitzman et al. (1995) used previous research to expand estimates of the cost of 17 major birth defects and cerebral palsy to the entire U.S. in 1992. Cost estimates from Waitzman et al. (1994) were adjusted to reflect national costs and expressed in 1992 dollars. The total combined cost of the conditions examined, accounting for the fact that some children suffered from multiple conditions, was estimated to be \$8 billion.

Chapter 3 Data Review

1. Introduction

Estimation of the economic values of the human reproductive and developmental impairment requires a range of empirical information. While it would be necessary to collect some data specifically for this purpose, a substantial amount of relevant information already is available. The government, largely through the National Center for Health Statistics, and private organizations maintain useful data sources. There is generally more information available related to the prevalence and cost of low birth weight babies and birth defects than infertility, partially due to the fact that the former end-points are more clearly defined.

The data review has been limited to sources providing information concerning the prevalence of the end-points under consideration. Estimation of economic values may also require information concerning various costs associated with the end-points. The majority of infertility treatment cost information has been summarized in the preceding literature review. Low birth weight and birth defects result in a range of costs occurring over the lifetime of the affected population. Because a wide range of data sources might be used depending on the types of costs to be included in an analysis of low birth weight or birth defects, cost related sources have not been reviewed.

2. Infertility

2.A. National Survey of Family Growth

The main data source related to the prevalence of infertility in the U.S. is the National Survey of Family Growth (NSFG). Almost all analyses involving the prevalence of infertility have relied on this series of surveys. Cycles I through IV of the NSFG were conducted in 1973, 1976, 1982 and 1988. In 1990, there was a telephone re-interview following cycle IV. The survey for cycle V was conducted in 1995 and first report based on this data is expected to be released in early 1997.

Cycle IV was based on a survey of 8,450 women drawn from households that had been interviewed for the National Health Interview Survey. All statistics are based on participants' responses rather than medical examinations or records.

Cycles I and II focused only on women aged 15-44 who were currently or previously married. Cycles III and IV expanded the survey population to women aged 15-44 of all marital statuses who were not institutionalized or in the military.

The NSFG contains a range of general demographic information as well as information specifically related to child-bearing. Of particular interest are the variables related to difficulty with child-bearing. The survey provides two current, not lifetime,

measures of difficulty with child-bearing. "Infertility" applies to married couples with wives 15-44 years of age who have had twelve or more months of intercourse without contraception and have not become pregnant. "Impaired fecundity" refers to women of all marital statuses for whom it is impossible to conceive or difficult or dangerous to carry to term.

The percent of married couples who are infertile is less than the percent with impaired fecundity, since infertility involves difficulty conceiving while impaired fecundity includes difficulty conceiving and difficulty or danger in carrying a pregnancy to term. Infertility was measured only for married couples while impaired fecundity was measured for both married and unmarried women.

While the fecundity status of an unmarried woman is determined by the status of the woman, the fecundity status of a married woman is determined by reproductive difficulties arising from either partner. Fecundity status is classified as: contraceptively sterile, surgically sterile for noncontraceptive reasons, nonsurgically sterile, long interval, subfecund and fecund. Nonsurgically sterile, long interval and subfecund are sometimes classified as impaired fecundity (Judkins, Mosher and Botman, 1991).

Numerous studies have used the NSFG to examine various issues surrounding child-bearing. Several are particularly relevant to an analysis of infertility treatment services. Henshaw and Orr (1987) have used the 1982 NSFG to estimate the number of women of reproductive age in need of infertility services. As indicated by the 1982 NSFG, 10.6 million (19.5%) women in the U.S. aged 15 to 44 were known fecund, 19.4 million (35.8%) were subfecund or infecund and 24.2 million (44.7%) were of unknown fecundity. 13.7 million (70.6%) of those who were infecund were surgically sterile. 3.5 million (18.1% of subfecund/infecund) could possibly be treated for their fecundity impairments; 2.4 million (68.9%) wanted more children. Of the 2.4 million women suffering from treatable fecundity impairments who wanted more children, 387,000 had received advice concerning infertility, 787,000 had received treatment and 1.2 million had received no services.

Wilcox and Mosher (1993) also examined the use of infertility treatment services as indicated by the 1988 NSFG. 2.3 million (42.9%) of women with impaired fecundity reported ever using infertility services. Advice was the most common service (25.4%); in vitro fertilization was used by 1.8%.

2.B. Royal Commission on New Reproductive Technologies

In reaction to the rapid pace of progress in the area of reproductive technologies and the scarcity of reliable information needed for health care policy decisions, the Canadian government created the Royal Commission on New Reproductive Technologies (RCNRT) in 1989. Following extensive literature reviews, surveys and analyses, the findings of the RCNRT were published as a fifteen volume set covering a wide range of topics related to reproductive technology. Topics covered included social values and

attitudes toward reproductive technologies (vol. 2), the prevalence of infertility (vol. 6), risk factors affecting fertility (vol. 7), and the cost-effectiveness of in vitro fertilization (vol. 11).

As part of the study, the RCNRT undertook new surveys and added questions to existing Canadian government surveys in order to gather a range of information related to fertility and reproductive technologies. Three surveys were conducted in 1991 and 1992 that sought to determine one and two year infertility prevalence among Canadian women aged 18-44 who were married or cohabiting for at least one or two years. Although the surveys were designed to be comparable to the NSFG, the RCNRT surveys differed from the NSFG in two main areas. While the NSFG defined infertility with respect to married women aged 15-44, the RCNRT included married and unmarried cohabiting women aged 18-44. When prevalence estimates were calculated to correspond with the definition of infertility provided by the NSFG, results between the two surveys were nearly identical (Dulberg and Stephens, 1993).

A second series of surveys sought to gain an understanding of people's attitudes toward infertility and reproductive technologies. Three surveys were structured so as to be representative of the overall population while two additional surveys focused specifically on cultural and religious influences on attitudes.

Through the use of focus groups, in-depth interviews, telephone interviews and phone surveys, the RCNRT elicited responses reflecting attitudes toward a range of topics related to infertility treatment. Several relevant topics include: family and children; assisted conception methods such as IVF, donor insemination and surrogacy; accessibility to assisted conception methods and payment for treatment (RCNRT, 1993).

To the extent that data used in the various studies are available, the RCNRT studies may provide valuable sources of information related to infertility and infertility treatment.

2.C. Other Estimates of the Prevalence of Infertility

Several other studies have attempted to estimate the prevalence of infertility. Batt et al. (1995), conducted a pilot study based on the Department of Motor Vehicles Registry for Upstate New York. The study sought to estimate the prevalence of self-reported pregnancy difficulty, to estimate the proportion of medically underserved couples and to identify reasons why couples do or do not seek treatment.

Respondents were asked if they ever had trouble becoming pregnant and if so for how long. Those having trouble were asked if they ever had seen a doctor and reasons why they had or had not sought assistance.

While infertility prevalence surveys other than the NSFG can add to the understanding of the magnitude of the problem of infertility, the statistical value of the

survey conducted by Batt et al. (1995) is limited by the small sample size of women reporting experiencing difficulty in becoming pregnant (n=39).

The infertility support group INCIID intends to conduct a survey designed to measure the degree of infertility and pregnancy loss among the general population with the aid of the on-line service Prodigy. The INCIID-Prodigy Infertility Survey will be able to sample from the population of 2 million Prodigy users.

2.D. Infertility Treatment Utilization

An estimate of the number of couples making use of several infertility treatments can be inferred from IVF registries. The Society for Assisted Reproductive Technology (SART) and the American Fertility Society (AFS) maintain a registry of assisted reproductive technology (ART) based on ART programs in the U.S., with a small number in Canada. In 1991, 212 programs reported initiating 33,000 cycles of ART treatment. There were 24,671 initiated IVF cycles, 5,452 initiated gamete intrafallopian transfer (GIFT) cycles and 2,104 initiated zygote intrafallopian transfer (ZIFT) cycles. 4,838 frozen embryo transfer (ET) procedures were also reported (SART 1993).

Data concerning IVF cycles is also maintained in France through the Fécondation In Vitro National (FIVNAT). Collection of data in the U.S. and France is based on voluntary reporting by IVF clinics in the respective countries. As indicated in Collins et al. (1995), the SART database contains about 95% of IVF cycles performed in the U.S. while FIVNAT includes about 80% of IVF services in France. Because reporting is voluntary, there may be low reporting rates by clinics with poor results.

Australia and the UK maintain databases concerning IVF treatments. Collins et al. (1995) note that publications concerning IVF in the UK and Australia report pregnancies and their outcomes but not the number of cycles performed per year. Reporting has become mandatory in the UK.

3. Low Birth Weight and Birth Defects¹⁶

There is a wide range of government data sources containing information related to low birth weight and birth defects. Vital records such as birth and infant death certificates currently record prenatal information and birth outcomes.¹⁷ In addition, surveys containing information related to childbirth have been conducted at various points in time. Some surveys, such as the National Natality Surveys and the National Maternal and Infant Health Survey, are termed followback surveys because survey participants are

¹⁶ Descriptions of National Center for Health Statistics datasets are largely derived from National Center for Health Statistics. 1994. "Catalog of Electronic Data Products," DHHS publication No. (PHS) 94-1213.

¹⁷ Birth and death certificates are collected by state governments. Information may not be uniformly reported by all states.

sampled from vital records. Followback surveys do not rely solely on the recollections of survey participants but also contain information from vital records. Finally, several government sponsored monitoring programs collect information related to birth weight and birth defects.

3.A. National Vital Statistics System - Natality

Natality vital statistics are based on the information reported in all birth certificates for a given year. Although natality statistics are collected by states, all states and the District of Columbia have participated in the aggregation of information since 1985. A revision of the United States Standard Certificate of Live Birth has led to some difference in the data reported before and after 1989. Prior to 1989, important medical variables contained on birth certificates included birth weight, gestation period and prenatal care. Demographic variables included residence of mother and age, education, marital status and race of mother and father. Medical variables from 1989 onward include: abnormal conditions of the newborn, alcohol use, birth weight, complications of labor or delivery, congenital anomalies, gestation period, medical risk factors, pregnancy history, prenatal care, tobacco use and weight gain during pregnancy. Similar demographic variables are reported as prior to 1989.

3.B. National Vital Statistics System - Fetal Death

Fetal death statistics are derived from fetal death reports registered with each state. Registration requirements for fetal deaths depend on the length of the gestation period and vary across states. Since 1989, medical and demographic variables reported on fetal death records are similar to those included in the natality statistics listed above.

3.C. National Vital Statistics System-Linked Birth-Infant Death

The linked birth-infant death dataset contains information on birth and death certificates for the entire population of infants that die in the U.S. each year. Information contained in the birth certificate has been described above. Additional information contained in the linked data set includes place of death, age at death and cause of death. 1991 is the most recent year for which the linked birth-infant death data set is available.

3.D. 1964-66, 1967-69, 1972 National Natality Survey (NNS)

The NNS contains data based on information from birth certificates and surveys of mothers. Each of the surveys contains a variety of socioeconomic variables such as education, income, household composition and mother's employment history. The 1967-69 surveys added information related to prenatal and postnatal care and cigarette smoking. The 1972 survey does not contain information related to cigarette smoking, but health variables based on questionnaires sent to physicians and hospitals were added.¹⁸ Health

¹⁸ Similar questionnaires were included in the 1980 National Natality Survey and the 1988 National Maternal and Infant Health Survey.

variables derived from the survey portion of the dataset include: prenatal and postnatal care, health of the infant and existence of maternal medical complications.

3.E. 1980 National Natality Survey and 1980 National Fetal Mortality Survey

The 1980 National Natality Survey and the 1980 National Fetal Mortality Survey, available in a combined form contain information gathered from birth and death certificates and surveys of the mothers and hospitals and physicians. The mother's questionnaire contains detailed information related to prenatal actions including cigarette smoking and alcohol consumption. The hospital and physician questionnaire asked detailed questions concerning the pregnancy history. Specific emphasis was placed on information related to radiation such as use of medical or dental x-rays or ultrasound.

3.F. 1988 National Maternal and Infant Health Survey (NMIHS)¹⁹

The NMIHS is a nationally representative study of mothers, prenatal care providers and hospitals based on a sample of 11,000 women who had live births, 6,000 who had infant deaths and 4,000 who had fetal deaths. Survey participants were sampled from the respective vital record systems.

Vital records information was supplemented by information gathered from a 35-page questionnaire mailed to mothers. The questionnaire included information on prenatal care, smoking, drinking and drug use, source of payment of prenatal care, gestation age and birth weight, child care and illness, and socioeconomic variables such as age, education, race and income.

During the course of the interview, mothers were asked to provide information related to prenatal care providers, hospitals used for delivery, and hospitals where the mother or child were admitted before or after delivery. Medical care providers identified by the mothers were sent questionnaires eliciting information related to prenatal care and medical utilization before, during and after the delivery.

The NMIHS is the equivalent of a combined National Natality Survey, National Infant Mortality Survey, and National Fetal Mortality Survey. As a result, the NMIHS can be used with these surveys to examine trends over time.

The NMIHS provides a range of information that can be used in analyzing factors influencing birth outcomes. Kogan et al. (1994), for example, have used the NMIHS to examine the impact of prenatal care as indicated by the U.S. Public Health Service Expert Panel on the Content of Prenatal Care on Birthweight. Sosniak et al. (1994) linked information contained in the NMIHS with the Environmental Protection Agency's

¹⁹For more information see: Sanderson M., P.J. Placek, and K.G. Keppel. 1991. "The 1988 National Maternal and Infant Health Survey: Design, Content, and Data Availability," *Birth*, 18(1), 26-32.

National Priorities List of hazardous waste sites in order to analyze the impact of proximity of residence to a hazardous waste site on birth outcome.

3.G. 1991 Longitudinal Follow-up to the National Maternal and Infant Health Survey

Women who were surveyed as part of the live birth cohort of the 1988 NMIHS were reinterviewed to obtain information related to child health and development, including the effects of low birth weight. Medical providers identified during the maternal survey were contacted in order to obtain information related to the type of health care organization and the child's health status and history. A follow-up survey was also conducted on the fetal and infant death cohorts of the 1988 NMIHS in order to obtain information related to respondents' health and subsequent fertility.

3.H. National Survey of Family Growth (NSFG)

While the NSFG was explained in greater detail above in terms of the information related to infertility, this survey also contains questions related to prenatal care, smoking and alcohol use during pregnancy.²⁰

3.I. National Health Interview Survey-Current Health Topics

The National Health Interview Survey is comprised of a core survey providing annual health information and supplementary surveys, the topics of which vary across years. Some of the relevant years and topics include:

1991: Pregnancy and Smoking, Child Health
1990: Pregnancy and Smoking
1988: Child Health
1985: Smoking History During Pregnancy
1981: Child Health Supplement

3.J. Birth Defects Monitoring Programs²¹

Information related to the occurrence of birth defects can be obtained from several birth defects monitoring programs. Programs differ by the method of data collection and the population monitored. *Passive* data collection relies on information obtained from birth certificates or hospital birth records. *Active* data collection involves hospital visits by researchers in order to obtain the necessary information directly. Because some birth defects are not observable immediately after birth, there is some level of error associated

²⁰For more information see: U.S. Department of Health and Human Services. 1995. "Health Aspects of Pregnancy and Childbirth: United States, 1982-88," Vital and Health Statistics, Series 23, No. 18.

²¹Information related to birth defects monitoring programs was obtained from *Teratology*, December 1993, 48(6) and conversations with Division of Birth Defects and Developmental Disabilities, National Center for Environmental Health, Centers for Disease Control and Prevention.

with the use of birth certificates as a source of birth defect information. As a result, active data is more reliable than passive data.

The Birth Defects Monitoring Program is a national surveillance program based on hospital discharge data from several thousand hospitals participating in the program. It was begun in 1974 with the aid of the March of Dimes and the National Institute of Child Health and Human Development. Since 1974, the Commission on Professional and Hospital Activities (CPHA) has processed information from approximately 1,200 hospitals participating in the program. For the years 1985-88, the McDonnell Douglas Health Information System (MDHIS) also provided information for the Birth Defects Monitoring Program.

Birth defects monitoring programs have also been established at the state level. About ten states have monitoring programs based on passive data collection. In addition, several state-level monitoring programs are based on active data collection, most notably the California Birth Defects Monitoring Program and the Metropolitan Atlanta Congenital Defects Program (MACDP). The Centers for Disease Control and Prevention, Emory University School of Medicine and the Georgia Mental Health Institute began the MACDP in 1967 in order to collect surveillance data for the metropolitan Atlanta area.

3.K. Pregnancy Risk Assessment Monitoring System (PRAMS)²²

PRAMS is a population based survey system used in thirteen states and the District of Columbia to monitor the effects of maternal behavior on birth outcomes. Information is obtained through a ten-page questionnaire containing both a standardized core and state specific questions sent to mothers sampled from birth certificate records. Topics in the core section include: prenatal care, maternal nutrition, obstetric history, use of alcohol and cigarettes, stress and economic status and infant status and health care.

PRAMS has been used to analyze elements of maternal behavior known to have an impact on birth outcomes. Bruce et al. (1993) examined self-reported maternal alcohol consumption prior to and during pregnancy and the magnitude of prenatal counseling about the effect of alcohol. Adams et al. (1992) utilized PRAMS to examine maternal smoking behavior prior to, during and following pregnancy and the relation between the source of prenatal care and maternal smoking.

²² For more information see: Adams, Melissa M., Holly B. Shulman, Carol Bruce, Carol Hogue, Donna Brogan and The PRAMS Working Group. 1991. "The Pregnancy Risk Assessment Monitoring System: Design, Questionnaire, Data Collection and Response Rates," *Paediatric and Perinatal Epidemiology*, **5**, 333-346.

Chapter 4

Recommendations for Phase II

1. Introduction

The following sections lay out suggestions for future research regarding the value placed on the reproductive and developmental process. Theoretical issues are discussed, research techniques are proposed and tasks needed to implement the techniques are outlined.

2. Infertility

Exposure to chemicals can have an impact on the human reproductive system in several ways. The reproductive system of an individual exposed to the chemical may be affected, limiting his or her ability to reproduce. Alternatively, exposure to certain chemicals prior to or during pregnancy may affect the development of the reproductive system of the fetus, leading to reproductive impairments later in life. Consequences of exposure thus occur in generations following the generation exposed to the chemicals. Much of the discussion of endocrine disruptors has focused on this second effect. Current theories suggest that exposure to chemicals altering the balance and function of hormones at critical points in the development of the fetus can have significant effects on the reproductive status of the child.

The benefits associated with a reduction in exposure to these chemicals depend on how the chemicals affect the reproductive process. Different values may be placed on current versus future fertility. The value placed on current fertility can be represented by the willingness to pay of potential parents for an increased probability of a successful pregnancy. Similarly, the value placed on future fertility can be estimated by the willingness to pay of parents for normal reproductive ability in their children.

Different techniques are required to estimate these different willingness to pay values. Estimation of the willingness to pay to reduce the probability that one's children will experience reproductive difficulties is limited to stated preference techniques. Estimation of the welfare change associated with a change in exposure to this set of chemicals is best estimated through methods such as conjoint analysis or contingent valuation surveys that elicit information concerning how much individuals would be willing to pay to reduce the probability that their children would suffer from reproductive impairments. In the sense that the conditions are long term and have large perceived costs, such a survey might be similar to stated preference methods used to determine willingness to pay to reduce the likelihood of low birth weight or birth defect births.

When estimating the willingness to pay to reduce current reproductive impairment, information based on couples' actions in addition to stated responses may be utilized. Expenditures by infertile couples on infertility treatments reflect the value that is placed on moving from a state of infertility to fertility. As a result of the broader range of available

estimation techniques, the following recommendations for future research focus on current fertility.

The following three sections outline research recommendations for an analysis of the valuation of reproductive ability. The first section contains an illustrative and preliminary microeconomic model that explains the demand for infertility treatment. The second section explains recommended approaches toward future research and the methods by which this may be accomplished. The final section lays out specific tasks.

2.A. Household Demand for Infertility Treatment

2.A1. Assumptions

It is assumed that some unspecified set of chemical compounds causes reproductive problems in humans of child-bearing age. That is, the problems manifest themselves in terms of reduction in the probability of child-bearing for couples between the ages of 15 and 44. Of particular concern is the welfare effects that these pollutants have on rates of *infertility*, where infertility is defined as the inability to conceive after 12 months of intercourse without contraception.

We examine the issues of child-bearing and infertility treatment at the *couple* (household) level. We assume there exists a couple utility function U , and that the male, female or both may be clinically affected by the compound(s) of interest. We define two states of the world with respect to couples' child-bearing -- *fertility* and *infertility*. Infertility is defined as above, i.e., "inability to conceive after 12 months of intercourse without contraception," while fertility is infertility's complement, i.e., successful conception within a 12-month period of intercourse. Each couple has a set of biological factors that determines whether it is fertile or infertile. These factors are distributed throughout the population of couples in a random manner. There exists a set of ambient chemical compounds that also affect fertility. These compounds are assumed to be uniformly distributed such that each couple is equally exposed. Each couple has its own tolerance to these chemicals. When the exposure of a fertile couple exceeds this tolerance, the couple becomes infertile (with certainty). Couples are ignorant of the compounds' presence and do not know whether infertility is due to exogenous biological factors, or to exposure to chemical compounds beyond the couple's tolerance.

Infertile couples may become fertile by expending effort on infertility treatments (effort is composed of both money and any number of nonpecuniary items including the couple's time). The effectiveness of infertility treatments to bring forth the fertility state is uncertain. With treatment, there is positive probability that a couple will move to the fertile state. Holding the couples' biological factors constant, the greater the treatment effort, the greater the probability of moving from the infertile to the fertile state.

Finally, each couple perceives the benefits (welfare gain) of unassisted (no treatment) conception and assisted (with treatment) conception to be equal.

2.A.2. Infertility Treatment Process

A couple suffering from infertility may undergo an episode of treatment in order to increase the likelihood of becoming fertile. If an episode of treatment is not successful, a couple may undergo an additional episode of the treatment or may select to undergo a different treatment. Treatment processes vary by success rates and costs. In addition, some treatments are more effective than others given the underlying cause of infertility.

Although success rates vary by the treatment procedure, it is not likely that a treatment will be successful after the first episode and many couples undergo multiple episodes of infertility treatment. The underlying probability of successful treatment depends on the biological attributes of the couple and the level of pollution exposure.

A couple that elects to begin infertility treatment is aware of the underlying probability of success in the general population but not to themselves. Due to this uncertainty, the perceived benefits and costs of treatment are important in determining whether or not treatment will result in a net benefit to the couple. Heterogeneous preferences for child-bearing result in different perceived benefits of treatment across couples. The overall cost of treatment depends on direct medical costs and indirect costs. Some indirect costs, such as the opportunity cost of the couple's time, can be objectively estimated based on wage rates and time estimates. Other indirect costs such as physical discomfort and psychological stress are more difficult to quantify and may change over time as the couple progresses through the treatment. For a risk-neutral couple, the overall cost of a successful pregnancy depends on the couple's perceived probability of becoming pregnant through treatment and the perceived direct and indirect costs of treatment. The decision to undergo and continue treatment depends on the couple's perceived net benefits of treatment.

Although medical consultation may provide an indication of the magnitudes of the various costs and the chance of success, a couple updates its perceptions of indirect costs and the probability of successful treatment as it progresses through the treatment process. Continued failure may be an indication that the couple is located at the lower end of the distribution of success probabilities. In addition, elements of some indirect costs can only be understood through experiencing the treatment. As a result, the estimated net benefit of treatment may change as the couple progresses through the treatment process, causing some couples to discontinue treatment.

2.A.3. A Stylized Model of the Demand for Infertility Treatment

The i th couple's fertility state is a function of couple's biological attributes, A_i , and the level of exposure to pollution, d_i . This can be represented by a fertility indicator function:

$$f_i = f(A_i, d_i)$$

where:

$$f_i = 1 \text{ if the couple is infertile}$$

$f_i = 0$ if the couple is fertile

An increased exposure to pollution or poor biological attributes increase the likelihood of a couple becoming infertile. A couple that is infertile can increase the likelihood of moving to the fertile state by undergoing an episode of infertility treatment, b_i^{tj} (where t, j and i indicate treatment, episode and couple, respectively). The fertility indicator function can thus be written as:

$$f_i = f(A_i, d_i, b_i^{tj})$$

The utility of the *i*th couple depends on a Hicksian composite of all commodities, X_i , leisure time, L_i , fertility state, f_i , and nonpecuniary factors (other than time)

associated with undergoing infertility treatment, s_i^{tj} .²³

$$U_i^{tj} = U(X_i, L_i, f_i, s_i^{tj})$$

Due to the uncertain outcome of infertility treatment, an infertile couple maximizes expected utility for episode *j* of treatment *t* subject to income and time constraints.

$$\text{Max } EU_i^{tj}(X_i, L_i, f_i, s_i^{tj})$$

$$\text{st } I_i + w_i^f(t_f - l_i^f - t_i^{bf}) + w_i^m(t_m - l_i^m - t_i^{bm}) = p_x X_i + p_b b_i^{tj}$$

$$T = t_f + t_m$$

$$\text{where } EU_i^{tj}(X_i, L_i, f_i, s_i^{tj}) = \delta_i^{tj} U_i^{tj}(X_i, L_i, 0, s_i^{tj}) + (1 - \delta_i^{tj}) U_i^{tj}(X_i, L_i, 1, s_i^{tj})^{24}$$

The first constraint indicates that the sum of nonwage income and the wage income of the husband and the wife equals spending on the Hicksian composite good and an episode of infertility treatment, $p_b b_i^{tj}$, where p_b , is the price of the treatment b_i^{tj} . The second constraint indicates that the total time available equals the time available to each spouse.

Expected utility is partially determined by δ_i^{tj} , the perceived probability of the current episode *j* of the current infertility treatment *t* leading to a successful outcome. δ_i^{tj} depends on both the couple's private expectations for success and the medical estimate of success. In addition, both the couple's and the medical probability estimates are conditional on previous experience (since biological attributes are heterogeneous, previous failures reveal information concerning a couples location along the distribution). Information can be learned both from previous episodes of the current treatment (such as previous cycles of IVF) and previous treatments (such as drugs which induce ovulation

²³ If no infertility treatment is undertaken $s_i^{tj} = 0$.

²⁴ If a couple does not undergo treatment $EU_i^{tj} = U_i(X_i, L_i, 1, 0)$.

employed as a treatment before IVF). Using Viscusi's prospective reference theory (Viscusi, 1989) this relation can be describe as:

$$\delta_i^{ij} = \left[\alpha_i (\delta_{ci}^{ij} | \theta_{ii}, \dots, \theta_{li}, \theta_{oi}) + \beta_i (\delta_m^{ij} | \theta_{ii}, \dots, \theta_{li}, \theta_{oi}) \right]$$

Where δ_{ci}^{ij} and δ_m^{ij} are the couple's and medical perceived probabilities and α_i and β_i are the weights given to each of these probabilities by the couple. Prior information includes expectations before any medical treatment has been undertaken, θ_{oi} , information from previous treatments, $\theta_{i-1,i}, \dots, \theta_{li}$, and information from previous episodes of the current treatment, θ_{ii} .

A couple will initiate treatment if the expected utility from the first treatment undergone, $EU_i^{1,1}$, is greater than the expected utility of no treatment, subject to the constraints:

$$\delta_i^{1,1} U_i^{1,1}(X_i, L_i, 0, s_i^{1,1}) + (1 - \delta_i^{1,1}) U_i^{1,1}(X_i, L_i, 1, s_i^{1,1}) > U_i(X_i, L_i, 1, 0)$$

A couple will undergo an additional episode of treatment if the expected utility is greater than the expected utility from undergoing a different treatment or dropping out of treatment:

$$\delta_i^{1,2} U_i^{1,2}(X_i, L_i, 0, s_i^{1,2}) + (1 - \delta_i^{1,2}) U_i^{1,2}(X_i, L_i, 1, s_i^{1,2}) > \delta_i^{2,1} U_i^{2,1}(X_i, L_i, 0, s_i^{2,1}) + (1 - \delta_i^{2,1}) U_i^{2,1}(X_i, L_i, 1, s_i^{2,1})$$

and

$$\delta_i^{1,2} U_i^{1,2}(X_i, L_i, 0, s_i^{1,2}) + (1 - \delta_i^{1,2}) U_i^{1,2}(X_i, L_i, 1, s_i^{1,2}) > U_i(X_i, L_i, 1, 0)$$

Similarly, a couple will switch to a new treatment if this alternative yields the highest expected utility:

$$\delta_i^{2,1} U_i^{2,1}(X_i, L_i, 0, s_i^{2,1}) + (1 - \delta_i^{2,1}) U_i^{2,1}(X_i, L_i, 1, s_i^{2,1}) > \delta_i^{1,2} U_i^{1,2}(X_i, L_i, 0, s_i^{1,2}) + (1 - \delta_i^{1,2}) U_i^{1,2}(X_i, L_i, 1, s_i^{1,2})$$

and

$$\delta_i^{2,1} U_i^{2,1}(X_i, L_i, 0, s_i^{2,1}) + (1 - \delta_i^{2,1}) U_i^{2,1}(X_i, L_i, 1, s_i^{2,1}) > U_i(X_i, L_i, 1, 0)$$

Since utility maximization will result in a couple purchasing or not purchasing one episode of infertility treatment at a point in time, couple i's demand function for an episode of treatment will not be continuous. Rather, a couple will undergo an episode of treatment if the cost of treatment is below the couple's reservation price for treatment, p_i^* .

If one assumes that there is sufficient variation in couples' reservation prices, aggregation of the individual utilization of a given infertility treatment yields a continuous market demand function for treatments. In addition to prices, p_x , nonwage and wage income $I_i + w_i^f(t_f - l_i^f - t_i^{bf}) + w_i^m(t_m - l_i^m - t_i^{bm})$, and the cost of treatment, $p_b b_i^{ij}$, demand for an infertility treatment is dependent on couples' perceptions of success rates and nonpecuniary costs.

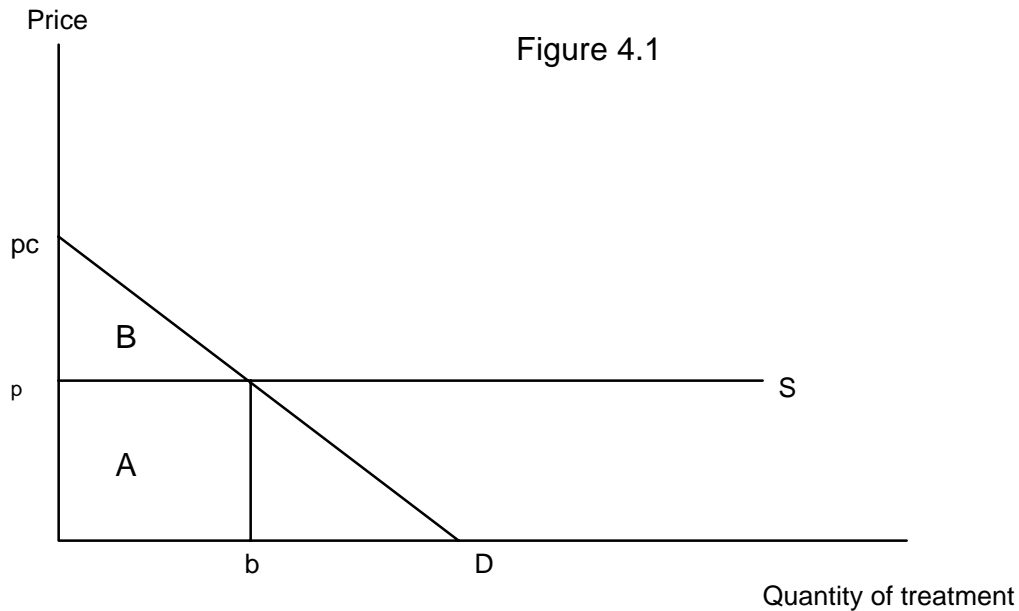
The market for all infertility treatments is characterized by a set of demand functions $b_t, t=1..T$,

$$b_t = b(p_x, I_i + w_i^f (t_f - l_i^f - t_i^{bf}) + w_i^m (t_m - l_i^m - t_i^{bm}), p_b b_i^{ij}, \theta_{ii}, \dots, \theta_{0i}, s_i^{ij}) \quad i=1..N,$$

where N indicates the number of infertile couples seeking treatment.

2.B. Research Approach

Figure 4.1 provides a simple graphical portrayal of the market for infertility treatment. The graph assumes a perfectly elastic supply (line segment pS) of infertility treatment services. We observe in the market a price for treatment equal to p and a quantity consumed b. Without any additional information, a lower bound on the value society places on these services is given by A. If we knew the choke price pc, we could assume a linear demand (given by the line segment pcD) and add to the value the consumer surplus triangle B.



If time and other nonpecuniary costs associated with treatment are large, then the price-quantity pair, pb, does not lie on the consumer's demand curve. Rather, the monetized value of these nonpecuniary costs must be added to p causing the "full" price of treatment to rise to p'. This is illustrated in figure 4.2. If we knew the monetized value of these nonpecuniary costs (the areas B+C) they could be added to the area A to more accurately capture the value of fertility. It is recommended that an estimation of the value of fertility not ignore these potentially significant nonpecuniary costs. Estimation of these costs might be obtained via a phone survey of couples currently undergoing or who have undergone infertility treatments. One objective of the survey would be to gather behavioral information with respect the time and other nonpecuniary costs of treatment.

Figure 2 reveals that the entire value of b units of infertility treatment is the area A+B+C+D. To obtain the area D an estimate of the choke price is required (in addition to a linearity assumption). The choke price is not a piece of observable information and can

be obtained only by stated preference approaches. As a result, the survey proposed above should also collect information needed for an estimate of the choke price so that a complete valuation of fertility treatments may be derived.

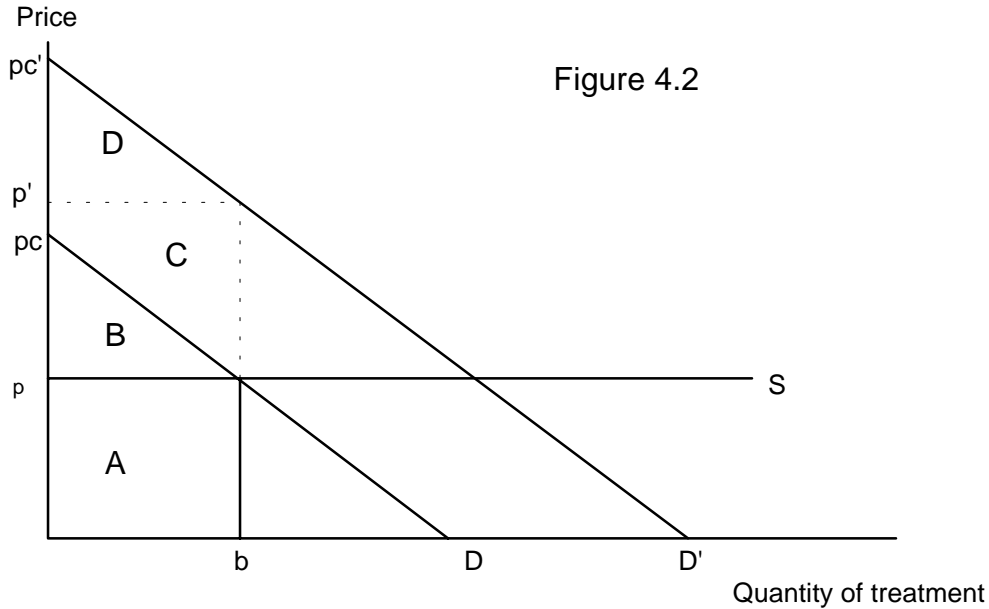


Figure 4.2

2.C. Tasks

2.C.1. Definition of Relevant Treatment Markets

Couples undergoing infertility treatment may choose from a variety of processes that vary according to success rates and costs in treating the underlying cause of infertility. An initial task of future research should be to clearly define the major treatment processes. This may be accomplished through a literature review as well as interviews of infertility treatment practitioners.²⁵ Emphasis should be placed on processes that are most effective in treating causes of infertility that might stem from exposure to pollution (e.g. male infertility).

2.C.2. Cost Estimation

As explained above, estimation of the value of fertility requires estimates of the market price of medical treatment, additional nonpecuniary costs, and the choke price at which treatment demand is reduced to zero. A portion of this information may be obtained from market data, where available. This data can be supplemented, to the extent possible, by information collected from those seeking and participating in treatments. Collection of market data (transactions) and unobserved behavioral data (time

²⁵ The OTA, for example, used the opinions of government and private sector infertility treatment experts to describe several infertility treatment processes (OTA, 1988). This information needs updating, however.

requirements, other nonpecuniary costs of treatment and the choke price) via direct questioning from the same sources provides the necessary information for the demand estimation process while ensuring that the observations are based on the same underlying preferences.

The source of survey participants would be determined by the degree of access to couples undergoing infertility treatment; possible sources include major infertility treatment centers and national infertility support organizations such as RESOLVE and the International Council on Infertility Information Dissemination (INCIID). Couples may also be sought (for information on nonpecuniary costs) from participants in the Canadian health care system who have used fertility services. Access to this population may be provided by consultants to the project, Dr. Bernie O'Brian and R. Goeree, Centre for Evaluation of Medicines, Father Sean O'Sullivan Research Centre, St. Joseph's Hospital, Ontario, Canada. Research into the effects of IVF on patient quality of life has been conducted by Dr. Goeree in Canada and could be drawn on for future research.²⁶

Although the exact content of the survey would be determined as the research evolves, the survey methodology should account for the major characteristics of the infertility treatment process outlined above. For example, the infertility treatment process is characterized by subjective valuations that are likely to change as couples progress through the treatment process. For this reason, the survey should include couples at different stages of the treatment process in order to obtain a representative sample of experiences.

- Estimation of Medical Cost

Future research may seek to obtain estimates of the medical costs of treatment as indicated by couples participating in the proposed survey (and from information on their co-payment percentages, if any) and from existing aggregate market data.

- Estimation of Non-Monetary Costs

While this report has indicated that the opportunity cost of couples' time and the cost of physical discomfort are two significant nonpecuniary costs of treatment, it will be necessary to obtain a more comprehensive identification of the major nonpecuniary costs of treatment. Infertility support groups might provide information related to the major physical and psychological difficulties surrounding treatment. Once these cost sources have been defined, a stated preference survey of couples undergoing treatment should be used to estimate the magnitude of the costs.

- Estimation of Choke Price

²⁶ See Goeree R., R. Labelle and J. Jarrell. 1993. "Public Preferences Toward an In Vitro Fertilization Program and the Effect of the Program on Patients' Quality of Life," in *New Reproductive Technologies and the Health Care System: The Case for Evidence-Based Medicine*, Ottawa: Ministry of Supply and Services, 601-25.

Within the same survey instrument, questions should be asked to estimate the price level that would cause participants to drop out of the treatment process. Since there are several elements of the overall cost of treatment, the survey may involve altering different cost elements. For example, a question on the choke price based on changes in the time requirements for treatment may be easier for subjects to accept than a question based on hypothetical increases in the direct medical cost. The use of focus groups to identify appropriate questions is recommended.

- Estimation of the Quantity of Treatments Performed

To the extent possible, future research can make use of existing data, such as the SART-ASRM registry described above, to estimate the quantity of infertility treatments performed. In the case of treatments for which there is no existing utilization information, utilization may be inferred from consultations with practitioners or from other data sources such as the National Center for Health Statistics' National Survey of Family Growth.

2.C.3. A Natural Experiment

To the extent appropriate and practical, examination of the behavior of Canadians seeking IVF services may provide valuable insight into the value of fertility. This service, up until very recently, had been provided under Canada's national health care system. However, this service has been generally de-listed in Ontario, meaning that, as is true for most people in the U.S., the costs of this service will have to be borne by those using it. With the price for this treatment changing from zero to \$10,000 or more, it may be possible to estimate at least a portion of the demand curve.

3. Low Birth Weight and Birth Defects

While the rates of infertility, low birth weight and birth defects may all be affected by exposure to certain pollutants, substantial differences exist with regard to the factors determining the proper valuation methodology. For example, while many infertile couples desire a successful pregnancy, few are willing to obtain this goal irrespective of cost. A portion of infertile couples choose not to incur the costs of treatment and remain childless or choose an alternative such as adoption while others drop out of treatment before obtaining a successful outcome. It is not likely, however, that a couple having a child that is low birth weight or suffering from birth defects will choose not to provide the needed treatment for that child. One would expect that the demand curves for treatment of low birth weight and birth defects are much more inelastic than the demand curves for infertility treatments.

The two sets of endpoints also differ with regard to the question of whose welfare is relevant to the analysis. While infertility can be modeled in terms of the effect on the welfare of a *couple*, birth defects and low birth weight have an impact on not only the parents but also the child. An analysis of the latter endpoints is thus best framed in terms

of the *family*. One might hypothesize that an analysis based at the family level is more likely to involve a complex interaction of preferences. Actions that might result in utility to one family member, such as consumption of alcohol during the pregnancy, can result in disutility to other family members in the form of increased likelihood of low birth weight.

A final distinction between low birth weight and birth defects and infertility stems from the fact that there are more known behavioral factors affecting the likelihood of occurrence of low birth weight or birth defects than infertility. The scientific community has established more clearly the link between actions such as smoking during pregnancy and birth weight than it has for actions affecting fertility.

As a result of the differences between the two sets of endpoints, different valuation techniques are appropriate for analyses of infertility and low birth weight and birth defects. As described above, one may take advantage of the fact that the demand for infertility treatment services is responsive to price in order to estimate the demand curve for infertility treatment and the corresponding welfare measure. The likely extreme inelasticity of the demand for treatment of low birth weight or birth defects prevents one from using a similar approach for these endpoints.²⁷ At the same time, the existence of actions affecting the probability of the occurrence of low birth weight and birth defects provides for an alternate approach toward value estimation.

The following three sections outline research recommendations for an analysis of the valuation of a reduction in the incidence of low birth weight or birth defects. The first section defines the proper concept of cost associated with a medical condition and discusses issues important to a model of the cost of low birth weight or birth defects. The second section explains recommended approaches toward future research and the methods by which this may be accomplished. The final section lays out specific tasks.

3.A. Cost of Low Birth Weight or Birth Defects

3.A.1. Definitions of Cost

The occurrence of low birth weight or birth defects results in a range of costs to those affected by the condition, varying in magnitude and ease of estimation. Costs such as direct medical costs can be observed from existing markets. Health care markets can be used to obtain information concerning elements of the cost of treatment such as medicine or hospitalization. Other indirect costs of illness are more difficult to determine but still can be estimated. The cost of time spent sick, for example, can be estimated in terms of foregone wages.

Cost estimates based on a summation of the direct and indirect medical costs only provide a lower bound estimate of the true cost of a condition. The total cost of a

²⁷ Specifically, it is unlikely that one could estimate a choke price for low birth weight or birth defect treatments through stated preference techniques.

condition is comprised of not only medical and time costs, but also the disutility due to the condition and the cost of resources used to reduce the likelihood of the occurrence of the condition. Medical conditions are associated with pain and suffering, the cost of which is not reflected in the direct medical cost. In the case of the endpoints under consideration, especially severe cases of birth defects, the disutility of both parents and child from the condition may be a substantial component of the overall cost. In addition, individuals may expend resources in order to lower the probability of the occurrence of an adverse condition. A policy may not alter the incidence of a condition but may still have benefits if it frees up resources that would otherwise be used in defensive or mitigating activities. Cost estimation techniques which do not account for disutility or defensive expenditures will underestimate the true cost.

The proper basis for cost used in cost-benefit analysis should be the cost a medical condition imposes on society. In many cases, the costs facing individuals will not equal the social cost. This is especially true for medical costs, where the existence of health insurance reduces the cost of treatment borne by the individual. Estimation of the benefits associated with reduced occurrences of low birth weight or birth defects should include not only private valuations of the reductions but also the benefits accruing to society not included in the private valuations.

3.B. Economic Model of Low Birth Weight and Birth Defects

As described above, the economic literature has recognized that the overall cost of a medical condition includes elements beyond the direct medical cost. Expenditures may be made on defensive actions that lessen the chance of occurrence of a condition while a condition that does occur imposes costs of time and disutility in addition to medical costs. This concept has been expressed formally in several microeconomic models. Harrington and Portney (1987), for example, derived expressions for each of these cost components from a model based on individual utility maximization. Willingness to pay for a reduction in pollution was found to be equal to the changes in the costs of defensive and mitigating actions, time spent ill and utility resulting from the change in pollution.

The applicability of existing models to an analysis of low birth weight or birth defects is limited by the existence of characteristics of low birth weight and birth defects that distinguish these conditions from other acute or chronic illness. Most significantly, low birth weight and birth defects are conditions that have a direct impact both on the lives of the parents and the affected child. Unlike existing analyses which have focused on the impact of an illness on an individual, analysis of low birth weight and birth defects should account for the fact that the welfare of several individuals is directly influenced by the conditions. In addition to the obvious costs incurred by the child, medical costs, lost time and emotional distress are all costs borne by parents for at least a portion of the child's life. In some cases, costs may be borne by the parents after the child has reached adulthood. A logical basis for an analysis of the cost of low birth weight and birth defects would thus be the family. Underlying a family-level model is the assumption that the utilities of family members can be aggregated into a family utility function. A family utility

function should account for the fact that actions that increase the utility of some family members may reduce the utility of others.

The use of family utility in a model of low birth weight or birth defects can better represent the relation between the factors influencing birth outcome and the outcome. Unlike existing models where defensive actions or exposure to pollution by the individual are assumed to alter the probability of that individual contracting an illness, the probability that a child will be born low birth weight or birth defect is influenced by factors affecting the parents, especially the mother. Existing models may explain defensive actions such as the use of bottled water by an individual to reduce the chance of water borne disease but they are less applicable to an explanation of actions such as the reduction in alcohol consumption by the mother during pregnancy to increase the probability that the child will be born healthy. A model of low birth weight or birth defects must account for the fact that actions by one component of the family unit affect the health outcome of another member by changing the probability of low birth weight or birth defects.

A theoretic model of low birth weight or birth defects must also account for the fact that these conditions have much longer lasting effects than acute illness. From the point of view of the affected individual, the effects of low birth weight and birth defects start at birth and may last a lifetime depending on the type and severity of the condition. This difference in duration may necessitate different modeling of cost elements. For example, acute illness analyses typically model the cost of time spent ill in terms of foregone wages from current employment. This may be less applicable to conditions where the entire career path may be altered as a result of the condition.

Since costs occur over an extended period of time, the timing of mitigating actions may have an effect on the lifetime cost of the condition. Lewit et al. (1995), for example, note that neonatal intensive care procedures and early intervention programs may result in reductions in costs later in life. While the timing of mitigating actions is not generally specified in models of acute illness, more explicit modeling may be needed in an analysis of low birth weight and birth defects.

A goal of phase II research should be to develop a rigorous economic model that specifically accounts for the factors unique to the valuation of reducing risks of low birth weight and birth defects. A model might be developed by assuming that family i derives utility from a numeraire good, X_i , time spent at leisure, L_i , and the health status of the child at birth, S_i :

$$U_i = U_i(X_i, L_i, S_i)$$

where $S_i = 1$ if the child is born low birth weight or with a birth defect

$S_i = 0$ otherwise.

The health status of the child at birth is a function of actions taken by the parents prior to and during the pregnancy, D_i , and by the level of exposure to environmental pollutants, P_i .

$$S_i = S_i(D_i, P_i)$$

A family with a child born low birth weight or birth defect will incur both monetary and time costs as a result of the condition. The medical cost to family *i* resulting from condition *j* might be expressed as:

$$M_{ij} = m_{ij}S_i(D_i, P_i)$$

The loss of time to family *i* from condition *j* might be expressed as:

$$B_{ij} = b_{ij}S_i(D_i, P_i)$$

The family chooses time spent at leisure and expenditures on the composite good and defensive activities so as to maximize utility subject to budget and time constraints.

$$\max_{L_i, X_i, D_i} U_{ij}(X_i, L_i, S_i)$$

$$\text{s.t. } I_i + w_i [T - L_i - b_{ij}S_i(D_i, P_i)] = X_i + D_i + m_{ij}S_i(D_i, P_i)$$

$$\text{and } T \geq L_i + b_{ij}S_i(D_i, P_i)$$

where I_i and w_i are nonlabor income and the wage rate for family *i*.

It is important to note that the above equations assume the existence of a family utility function and do not specify what is included in the monetary and time costs, m_{ij} and b_{ij} . As a result, they should be viewed as suggestions of how a model might be developed rather than as a definitive basis for future analysis.

3.C. Research Approach

One method of estimating the value of a healthy birth infers the value from actions taken by the parents. Costs that individuals choose to incur in order to reduce the probability of an undesired outcome provide information related to preferences toward the outcome. Previous studies have examined the use of smoke detectors and seatbelts to infer the value associated with a reduction in the risk of death (Blomquist, 1979; Dardis, 1980). A range of actions that may be taken by parents during the pregnancy, such as eliminating cigarette, drug or alcohol consumption or use of prenatal care services, affect the probability that the baby will be born healthy. Information concerning the costs and the changes in the probability of a healthy birth can be used to estimate the value of a healthy birth.

The use of this technique requires knowledge of the perceived costs and effects of the actions taken. Some cost elements such as the medical cost of prenatal care can be

observed from market data. Other cost elements, such as the loss in utility associated with a reduction in cigarette use, are subjective in nature and vary across individuals. Similarly, an individual's perception of the impact of an action on the probability of a healthy birth may differ significantly from the views of the medical community. Since it is the individual's perceptions of the costs and effectiveness of the actions that are incorporated into the decision process, it is important to base value estimates on these variables.

A value estimate obtained in this manner will reflect the preferences of those from whom information was collected. To the extent that tastes and perceptions toward risk are heterogeneous, the value estimate may differ from the value of the underlying population. One might expect, for example, that perceptions toward risk might differ among those who stopped smoking at the start of pregnancy, those who did not stop and those who never smoked.

Alternately, stated preference methods such as contingent valuation or conjoint analysis can be used to determine individuals' willingness to pay to lessen the likelihood of occurrence of low birth weight or birth defects. The amount that an individual would be willing to pay to decrease the probability of occurrence of the conditions would be influenced by the total cost of the conditions incurred by the individual. As a result, contingent valuation or conjoint analysis would provide more accurate estimates of private value than a cost of illness analysis.

Previous studies have employed stated preference techniques to determine willingness to pay for various health outcomes. Viscusi, Magat, Huber (1991) and Krupnick and Cropper (1992) determined willingness to pay to reduce the probability of chronic bronchitis. A small group of studies have also been reported within the health literature addressing the application of contingent valuation to conditions such as chronic arthritis (Thompson, Read, Liang, 1984) and hypertension (Johannesson, Jönsson, Borgquist, 1991). These studies can provide some guidance in determining elements of survey design pertinent to health outcome analysis. For example, due to the large cost associated with low birth weight and birth defects, willingness to pay questions may be better framed in terms of changes of probabilities of occurrence rather than definite outcomes. Survey participants may be able to conceptualize an amount that they would be willing to pay for a slight reduction in the probability of low birth weight or birth defects but may have more difficulty estimating how much they would be willing to pay to prevent their children being born with one of the conditions with certainty. While previous analyses may provide some insight into survey design, care must be taken to account for the factors that distinguish low birth weight or birth defects from the conditions examined in these studies.

An analysis based on stated preference or revealed preference will reflect the private willingness to pay to reduce the likelihood of a low birth weight or birth defect birth. Estimation of the added social cost not paid by individuals will also be necessary in order to estimate the total cost of the endpoints.

3.D. Tasks

3.D.1. Development of Economic Model

Due to the lack of existing models that are applicable to an analysis of low birth weight or birth defects, an initial task of future research should involve the development of a formal model based on microeconomic principles. Such a model should describe the costs associated with the endpoints based on utility maximizing behavior at the family level. Existing economic models of morbidity or mortality may provide some guidance with the model development.

3.D.2. Estimation of Private Willingness to Pay

Stated preference techniques and observation of actions that have an impact on the birth outcome both can provide estimates of the private willingness to pay to increase the probability of a healthy birth. Since both techniques require survey data, a single survey might be used to collect information needed for each of the techniques in order to compare willingness to pay estimates between the methods.

3.D.3. Stated Preference

Estimation of the private willingness to pay to avoid the risk of low birth weight or birth defects could be obtained through stated preference techniques such as contingent valuation or conjoint analysis. The exact format and content of the survey would be determined as the research evolves, but guidance may be provided by existing studies that have considered elements of the health care field that differentiate it from other areas where stated preference techniques have been used (Gafni, 1991).

3.D.3.1 Actions Affecting Birth Outcome

- Determine actions affecting birth outcome

It will be necessary to determine what actions are perceived to have an impact on the health status of the child. Some actions, such as alcohol, tobacco, or drug consumption or use of prenatal care, have been mentioned above. The medical literature can be surveyed to develop a more comprehensive list.

- Determine perceived costs and impacts of actions

A survey should be conducted to determine the perceived costs of the actions taken to increase the likelihood of a healthy birth. The structure of such a survey may be patterned after existing research that has estimated willingness to pay based on perceived costs and impacts of actions. Chestnut et al. (1996), for example, estimated willingness to pay for changes in angina symptoms based on an averting behavior study that incorporated the perceived impacts of actions taken by respondents on the occurrence of the symptoms.

- Determine applicability of results

Analysis should be performed to determine if the perceptions of those who have taken actions differ from those of the medical community or general population.

3.D.4. Estimation of Additional Social Cost

It is necessary to determine what portion of the overall cost is not incurred by the family and thus would not be incorporated in the private willingness to pay to avoid the illness. Previous analyses may be used as guidance. For example, Cropper and Krupnick (1990) focused on the medical costs and lost productivity associated with chronic heart and lung diseases when estimating the social costs of these diseases.

Note: A more detailed description of Phase II recommendations related to low birth weight and birth defects will be provided in the future.

- Adamowicz, W., J. Louviere and M. Williams. 1994. "Combining Revealed and Stated Preference Methods for Valuing Environmental Amenities," *Journal of Environmental Economics and Management*, **26**, 271-292.
- Adams, Melissa M., Donna J. Brogan, Juliette S. Kendrick, Holly B. Shulman, Christine Zahniser, F. Carol Bruce and the PRAMS Working Group. 1992. "Smoking, Pregnancy, and Source of Prenatal Care: Results From the Pregnancy Risk Assessment Monitoring System," *Obstetrics & Gynecology*, **80** (5), 738-744.
- Adams, Melissa M., Holly B. Shulman, Carol Bruce, Carol Hogue, Donna Brogan and the PRAMS Working Group. 1991. "The Pregnancy Risk Assessment Monitoring System: Design, Questionnaire, Data Collection and Response Rates," *Paediatric and Perinatal Epidemiology*, **5**, 333-346.
- Auger, J., J.M. Kunstmann, F. Czyglik and P. Jouannet. 1995. "Decline in Semen Quality Among Fertile Men in Paris During the Past Twenty Years," *New England Journal of Medicine*, **332** (5), 281-285.
- Bartels, D. 1993. "The Financial Costs of In Vitro Fertilization: An Example from Australia," in P. Stephenson and M. Wagner, eds., *Tough Choices*, Philadelphia: Temple University Press.
- Batt, Ronald, Germaine Buck, Mark Baptiste et al. 1995. "A Population-Based Study of Infertile Women Who Do and Do Not Seek Services from Physicians," paper presented at the 51st Annual Meeting of the American Society for Reproductive Medicine, Seattle, Washington.
- Becker, Gary. 1960. "An Economic Analysis of Fertility," in *Demographic and Economic Change in Developed Countries*, Princeton, NJ: Princeton University Press and the National Bureau of Economic Research.
- Bleichrodt, H. and M. Johannesson. 1996. "The Validity of QALYs: An Experimental Test of Constant Proportional Trade-Off and Utility Independence," *Medical Decision Making*, **16** (4).
- Blomquist, Glenn. 1979. "Value of Life Savings: Implications of Consumption Activity," *Journal of Political Economy*, **87**, 540-558.
- Bruce, F. Carol, Melissa M. Adams, Holly B. Shulman, M. Louise Martin and the PRAMS Working Group. 1993. "Alcohol Use Before and During Pregnancy," *American Journal of Preventive Medicine*, **9** (5), 267-273.
- Cameron, Trudy Ann. 1992. "Combining Contingent Valuation and Travel Cost Data for the Valuation of Nonmarket Goods," *Land Economics*, **68** (3), 302-317.
- Carlsen E., A. Giwercman, N. Keiding and N. Skakkebaek. 1992. "Evidence for Decreasing Quality of Semen during the Past 50 Years," *British Medical Journal*, **305**, 609-613.
- Chaikind, S. and H. Corman. 1991. "The Impact of Low Birthweight on Special Education Costs," *Journal of Health Economics*, **10** (3), 291-311.

- Chestnut, Lauraine G., L. Robin Keller, William E. Lambert and Robert D. Rowe. 1996. "Measuring Heart Patients' Willingness to Pay for Changes in Angina Symptoms," *Medical Decision Making*, **16**, 65-77.
- Chu, R.C. 1988. *1985 Indirect Costs of Infant Mortality and Low Birth Weight*. Washington, DC: National Commission to Prevent Infant Mortality.
- Colborn, T., J. Myers and D. Dumanoski. 1996. *Our Stolen Future; How We Are Threatening Our Fertility, Intelligence, and Survival; A Scientific Detective Story*, New York: Dutton.
- Collins, John A. 1994. "Reproductive Technology-The Price of Progress," *New England Journal of Medicine*, **331** (4), 270-271.
- Collins, John A., Maria Bustillo, Robert D. Visscher and Lynne D. Lawrence. 1995. "An Estimate of the Cost of In Vitro Fertilization Services in the United States in 1995," *Fertility and Sterility*, **64** (3), 538-545.
- Cooper, Glinda S. 1986. "An Analysis of the Costs of Infertility Treatment," *American Journal of Public Health*, **76** (8), 1018-1019.
- Corman, H. 1994. "The Effects of Low Birth Weight and Other Medical Risk Factors on Resource Utilization in the Preschool Years," Report to the Center for the Future of Children.
- Corman, H. and S. Chaikind. 1993. "The Effect of Low Birthweight on the Health, Behavior, and School Performance of School-Aged Children," Working Paper No. 4409. Cambridge, MA: National Bureau of Economic Research.
- Cropper, Maureen L. and Alan J. Krupnick. 1990. "The Social Costs of Chronic Heart and Lung Disease," Resources for the Future Discussion Paper QE89-16-REV.
- Dardis, Rachel. 1980. "The Value of a Life: New Evidence from the Marketplace," *American Economic Review*, **70**, 1077-1082.
- Dulberg, Corinne S. and Thomas Stephens. 1993. "The Prevalence of Infertility in Canada, 1991-1992: Analysis of Three National Surveys," in *The Prevalence of Infertility in Canada*, Ottawa: Ministry of Supply and Services, 61-106.
- Eom, Y.S. and V.K. Smith. 1994. "Calibrated Nonmarket Valuation," Resources for the Future Discussion Paper 94-21.
- Erickson, J. D. 1993. "Introduction," *Teratology*, **48**: 545-546.
- Gafni, A. 1991. "Willingness-to-Pay as A Measure of Benefits," *Medical Care*, **29**, 1246-1252.
- Gafni, A. and S. Birch. 1995. "Preferences for Outcomes in Economic Evaluation: An Economic Approach to Addressing Economic Problems," *Social Science and Medicine*, **40** (6), 767-776.
- Gafni A., S. Birch and A. Mehrez. 1993. "Economics, Health, and Health Economics: HYE's Versus QALYs," *Journal of Health Economics*, **11**, 325-39.

- Goeree, Ron, Roberta Labelle and John Jarrell. 1993a. "Cost-Effectiveness of an In Vitro Fertilization Program and the Costs of Associated Hospitalizations and Other Infertility Treatments," in *New Reproductive Technologies and the Health Care System: The Case for Evidence-Based Medicine*, Ottawa: Ministry of Supply and Services, 569-599.
- Goeree, Ron, Roberta Labelle and John Jarrell. 1993b. "Public Preferences Toward an In Vitro Fertilization Program and the Effect of the Program on Patients' Quality of Life," in *New Reproductive Technologies and the Health Care System: The Case for Evidence-Based Medicine*, Ottawa: Ministry of Supply and Services, 601-625.
- Goldfarb, James M., Cynthia Austin, Hanna Lisbona, Barry Peskin and Marge Clapp. 1996. "Cost-Effectiveness of In Vitro Fertilization, *Obstetrics & Gynecology*, **87** (1), 18-21.
- Granberg, Maria, Matts Wikland, Lars Nilsson and Lars Hamberger. 1995. "Couples' Willingness to Pay for IVF/ET," *Acta Obstetrica et Gynecologica Scandinavica*, **74**, 199-202.
- Haan, Ger. 1991. "Effects and Costs of In-Vitro Fertilization: Again, Let's Be Honest," *International Journal of Technology Assessment in Health Care*, **7**, 585-593.
- Harrington, W. and M. Toman. 1994. "Methods for Estimating the Economic Value of Human Health Benefits from Environmental Improvement," Resources for the Future Discussion Paper 94-41.
- Harrington, W. and P. Portney. 1987. "Valuing the Benefits of Health and Safety Regulation," *Journal of Urban Economics*, **22**, 101-112.
- Henshaw, S.K. and M.T. Orr. 1987. "The Need and Unmet Need for Infertility Services in the United States," *Family Planning Perspectives*, **19**, 180-186.
- Hornberger, John C., Donald A. Redelmeier and Jeffrey Petersen. 1992. "Variability Among Methods to Assess Patients' Well-Being and Consequent Effect on A Cost-Effectiveness Analysis," *Journal of Clinical Epidemiology*, **45** (5), 505-512.
- Johannesson, M. 1995. "Quality-Adjusted Life-Years Versus Healthy-Years Equivalent-A Comment," *Journal of Health Economics*, **14** (1), 9-16.
- Johannesson, M. and B. Jönsson. 1991. "Economic Evaluation in Health Care: Is There a Role for Cost-Benefit Analysis?" *Health Policy*, **17**, 1-23.
- Johannesson, M., B. Jönsson and L. Borgquist. 1991. "Willingness to Pay for Antihypertensive Therapy: Results of A Swedish Pilot Study," *Journal of Health Economics*, **10**, 461-474.
- Johansson, Per-Olov. 1995. *Evaluating Health Risks: An Economic Approach*, Cambridge: Cambridge University Press.
- Joyce, T., M. Grossman and F. Goldman. 1986. "An Assessment of the Benefits of Air Pollution Control: The Case of Infant Death," Working Paper No. 1928. Cambridge, MA: National Bureau of Economic Research.

- Judkins, D.R., W.D. Mosher and S. Botman. 1991. "National Survey of Family Growth: Design, Estimation, and Inference," Vital and Health Statistics, Series 2, No. 109, National Center for Health Statistics, DHHS publication No. 91-1386.
- Kogan M.D., G.R. Alexander, M. Kotelchuck, and D.A. Nagey. 1994. "Relation of the Content of Prenatal Care to the Risk of Low Birth Weight. Maternal Reports of Health Behavior Advice and Initial Prenatal Care Procedures," *JAMA*, **271** (17), 1340-1345.
- Kopp, Raymond J. 1993. "Environmental Economics: Not Dead But Thriving," *Resources* (Spring), Resources for the Future, Washington, D.C.
- Krupnick, A.J. and M.L. Cropper. 1992. "The Effects of Information on Health Risk Valuations," *Journal of Risk and Uncertainty*, **5** (2), 29-40.
- Leibowitz, Arleen. 1990. "The Response of Births to Changes in Health Care Costs," *Journal of Human Resources*, **25** (4), 697-711.
- Lewit, Eugene M., Linda Schuurmann Baker, Hope Corman and Patricia H. Shiono. 1995. "The Direct Cost of Low Birth Weight," *The Future of Children*, **5** (1), 35-56.
- Lindert, Peter H. 1978. *Fertility and Scarcity in America*, Princeton, NJ: Princeton University Press.
- Loomes, Graham. 1995. "The Myth of the HYE," *Journal of Health Economics*, **14** (1), 1-8.
- Loomes, Graham and Lynda McKenzie. 1989. "The Use of QALYs in Health Care Decision Making," *Social Science and Medicine*, **28** (4), 299-308.
- Mehrez, A. and A. Gafni. 1989. "Quality-Adjusted Life-Years, Utility Theory, and Healthy-Years-Equivalents," *Medical Decision Making*, **9**, 142-149.
- Mitchell, R. M. and R. T. Carson. 1989. *Using Surveys to Value Public Goods: The Contingent Valuation Method*, Washington, DC: Resources for the Future.
- Mosher, W. D., and W. F. Pratt. 1990. "Fecundity and Infertility in the United States, 1965-88," *Advance Data*, Number 192, National Center for Health Statistics, U.S. Department of Health and Human Services, Public Health Service, December 4.
- Neumann, Peter J., Soheyla Gharib and Milton Weinstein. 1994. "The Cost of a Successful Delivery with In Vitro Fertilization," *New England Journal of Medicine*, **331** (4), 239-243.
- Neumann, Peter J. and Magnus Johannesson. 1994. "Willingness to Pay for In Vitro Fertilization: A Pilot Test Using Contingent Valuation," *Medical Care*, **32** (7), 686-699.
- Page, H. 1989. "Economic Appraisal of In Vitro Fertilization: Discussion Paper," *Journal of the Royal Society of Medicine*, **82**, 99-102.
- Paneth, Nigel. 1995. "The Problem of Low Birth Weight," *The Future of Children*, **5** (1), 19-34.

- Pliskin, J.S., D.S. Shepard and M.C. Weinstein. 1980. "Utility Functions for Life Years and Health Status," *Operations Research*, **28**, 206-224.
- Pratt, Donna, Burton F. Vanderlaan, Alan Dudkiewicz, Vishvanath Karande and Arnold L. Widen. 1994. "A Managed Care Provider's Approach Toward Mandated Infertility Coverage: The Illinois Family Building Act," *Journal of Assisted Reproduction and Genetics*, **11** (9), 433-438.
- Royal Commission on New Reproductive Technologies. 1993. "An Overview of Findings in This Volume," in *Social Values and Attitudes Surrounding New Reproductive Technologies*, Ottawa: Ministry of Supply and Services, 1-34.
- Ryan, Mandy. 1994. "Using Economics to Assess the Benefits of Assisted Reproductive Techniques," Health Economics Research Unit Discussion Paper 04/94, Departments of Public Health and Economics, University of Aberdeen, Aberdeen.
- Sagoff, M. 1993. "Environmental Economics: An Epitaph," *Resources* (Spring), Resources for the Future, Washington, D.C..
- Sanderson, M., P.J. Placek and K.G. Keppel. 1991. "The 1988 National Maternal and Infant Health Survey: Design, Content, and Data Availability," *Birth*, **18** (1), 26-32.
- Scanlon, T. M. 1991. "The Moral Basis of Interpersonal Comparisons," in J. Elster and J. E. Roemer, eds., *Interpersonal Comparisons of Well-Being*. Cambridge: Cambridge University Press.
- Schenker, J. 1993. "Medically Assisted Conception: The State of the Art in Clinical Practice," in P. Stephenson and M. Wagner, eds., *Tough Choices*, Philadelphia: Temple University Press.
- Schwartz, R.M. 1989. "What Price Prematurity?" *Family Planning Perspectives*, **21** (4), 170-174.
- Shiono, Patricia H. and Richard E. Behrman. 1995. "Low Birth Weight: Analysis and Recommendations," *The Future of Children*, **5** (1), 4-18.
- Society for Assisted Reproductive Technology, American Fertility Society. 1993. "Assisted Reproductive Technology in the United States and Canada: 1991 Results from the Society for Assisted Reproductive Technology Generated from the American Fertility Society Registry," *Fertility and Sterility*, **59** (9), 56-62.
- Sosniak, W.A., W.E. Kaye and T.M. Gomez. 1994. "Data Linkage to Explore the Risk of Low Birthweight Associated with Maternal Proximity to Hazardous Waste Sites from the National Priorities List," *Archives of Environmental Health*, **49** (4), 251-255.
- Stern, Zvi, Neri Laufer, Rami Levy, Daniella Ben-Shushan and Shlomo Mor-Yosef. 1995. "Cost Analysis of In Vitro Fertilization," *Israel Journal of Medical Sciences*, **31** (8), 492-496.
- Thompson, M.S., J.L. Read and M. Liang. 1984. "Feasibility of Willingness-to-Pay Measures in Chronic Arthritis," *Medical Decision Making*, **4**, 195-215.

- Torrance, George W. and David Feeny. 1989. "Utilities and Quality-Adjusted Life Years," *International Journal of Technology Assessment in Health Care*, **5**, 559-575.
- U.S. Congress, Office of Technology Assessment. 1988. *Infertility: Medical and Social Choices*. Washington, DC: U.S. Government Printing Office.
- U.S. Department of Health and Human Services. 1994. "Catalog of Electronic Data Products." DHHS Publication No. (OHS) 94-1213.
- U.S. Department of Health and Human Services. 1995. "Health Aspects of Pregnancy and Childbirth: United States, 1982-88," *Vital and Health Statistics, Series 23*, No. 18.
- Van Voorhis, Bradley J., Craig H. Syrop, Brian D. Allen, Amy E.T. Sparks and Dale W. Stovall. 1995. "The Efficacy and Cost Effectiveness of Embryo Cryopreservation Compared with Other Assisted Reproductive Techniques," *Fertility and Sterility*, **64**, 647-650.
- Viscusi, W.K. 1989. "Prospective Reference Theory: Toward an Explanation of the Paradoxes," *Journal of Risk and Uncertainty*, **2**, 235-264.
- Viscusi, W.K., W.A. Magat and J. Huber. 1991. "Pricing Environmental Health Risks: Survey Assessments of Risk-Risk and Risk-Dollar Trade-offs for Chronic Bronchitis," *Journal of Environmental Economics and Management*, **21**, 32-51.
- Wagner, M. and P. Stephenson. 1993. "Infertility and In Vitro Fertilization: Is the Tail Wagging the Dog?" in P. Stephenson and M. Wagner, eds., *Tough Choices*, Philadelphia: Temple University Press.
- Waitzman, Norman J., Patrick S. Romano and Richard M. Scheffler. 1994. "Estimates of the Economic Costs of Birth Defects," *Inquiry*, **33** (Summer), 188-205.
- Waitzman, N.J., P.S. Romano, R.M. Scheffler and J.A. Harris. 1995. "Economic Costs of Birth Defects and Cerebral Palsy-United States, 1992," *Morbidity and Mortality Weekly Report*, **44** (37), 694-695.
- Wakker, Peter. 1996. "A Criticism of Healthy-Years Equivalents," *Medical Decision Making*, **16** (3), 207-214.
- Weinstein, M. and J. Pliskin. 1996. "HYEs: What Are the Issues?" *Medical Decision Making*, **16** (3), 205.
- Wilcox, Lynne S. and William D. Mosher. 1993. "Use of Infertility Services in the United States," *Obstetrics and Gynecology*, **82** (1), 122-127.
- Williams, B. A. O. 1985. *Ethics and the Limits of Philosophy*. London: Collins/Fontana Press.
- Willis, Robert J. 1973. "A New Approach to the Economic Theory of Fertility Behavior," *Journal of Political Economy*, **81** (2), S14-S64.

Wolpin, Kenneth. 1984. "An Estimable Dynamic Stochastic Model of Fertility and Child Mortality," *Journal of Political Economy*, **92** (5), 852-874.