

Improving Estimates of Caregiver Time Cost and Family Impact Associated with Birth Defects

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ABSTRACT

Background: Birth defects impose substantial costs on both families and society because of medical, developmental, and special education needs. Caring for children with birth defects also may influence caregiver time and impact the family. However, the economic cost of caregiver time and other impacts on the family has received far less attention than traditional health-care costs.

Methods: This study reviews the literature on measuring caregiver time costs and family impact in an economic framework. The economic framework involves translating caregiver time or difficulties into appropriate units such as cost or quality adjusted life years (QALYs).

Results: Despite the potential important contribution of caregiver time costs to the total cost estimate of birth defects, few studies estimate caregiver time costs related specifically to birth defects. Only two studies provide estimates of these costs. Recent work has investigated the impact of chronic illness on caregivers in QALY terms, but birth defects have not been studied. Several issues need to be addressed in both the estimation of caregiver time costs and family impact to improve cost estimates.

Conclusions: Improved estimates of caregiver time costs and impact on the family will assist policy makers in allocating resources for the prevention and treatment of birth defects. Future research should investigate the economic costs of caregiver time and family impact associated with caring for children with birth defects.

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costs should be included in any benefit assessment, few studies estimate the caregiver time costs associated with birth defects, and several issues must be addressed to translate family impact into economic values.

We reviewed the literature on caregiver time costs and family impact in an economic framework. We used an economic framework because of the increasing use of cost-benefit and cost-effectiveness analyses to guide policy decisions for allocating resources to birth defect treatment and prevention programs. Including relevant information about caregiver time costs or family impact in these calculations could improve resource allocations for the prevention and treatment of birth defects.

The article has three major sections. The first section addresses the calculation of caregiver time costs, the second section describes potential methods for measuring family impact, and the final section illustrates the use of this information to guide policy on the allocation of public funds for birth defect treatment and prevention strategies.

MEASUREMENT OF CAREGIVER TIME COSTS

The measurement of caregiver time costs should follow general principles of cost-of-illness methodology. Cost-of-illness methodology requires the analyst to consider whether the results will be used for prevention or treatment studies and to assess the incremental costs associated with the program (Hartunian et al., '80). For prevention studies, an incidence approach is relevant and typically provides an estimate of the cost per new case. For example, recent estimates of the cost of the major birth defects provides policy makers with an estimate of the medical, developmental, and special education costs that could be saved by preventing a single birth defect (Waitzman et al., '96). This approach uses per capita costs among a prevalent population to

INTRODUCTION

Estimates of the cost of medical, developmental, and special education services for children with major structural birth defects can be used to determine the potential benefits of birth defect prevention programs (Waitzman et al., '96). These benefit calculations, however, often exclude the caregiver time costs or family impact associated with birth defects. Although these

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estimate costs for an incident cohort, discounted back to present value terms. An incidence approach also is relevant for estimating caregiver time costs, but several assumptions are required to estimate such costs using this approach.

Cost analysis typically assesses the incremental costs associated with birth defects. This method estimates the medical, developmental, or special education costs for a child with a birth defect in excess of those costs for the average child. The same is true for consideration of caregiver time costs. The working assumption is that the birth defect increases the time commitment of the caregiver in excess of the average child (Crowe, '93; Clipp and Moore, '95; Barnett and Boyce, '95; Moore and Clipp, '94)

If a child with a birth defect requires additional caregiver time to aid with activities of daily living, the increased time commitment is an expense because it reduces the caregiver time available for other activities, including work and leisure. Valuing the opportunity cost of these other activities, however, often proves difficult. The current recommendation is to consider only differences in work time using a wage rate to value an hour of work time (Luce et al., '96). This too can prove difficult if people do not work for a wage.

Studies have shown reductions in work time for parents of children with chronic health conditions (Salkever, '85; Salkever, '82; Breslau, '83; Breslau and Salkever, '81). Although useful for documenting the impact of chronic illness on caregiver time, these estimates provide little guidance for estimating caregiver time costs associated with a *specific* birth defect. Time costs were reported for specific birth defects in two studies: spina bifida and Down syndrome.

The study of caregiver time costs associated with spina bifida reported average reductions of 14 hours per week in paid work time for mothers and 5 hours per week for fathers (Lipscomb, '86). A much smaller reduction in work time was reported in the study of caregivers of children with Down syndrome. In this study, paid work time was reduced by 6.6 hours per week for mothers, but there was no difference in paid work time for fathers (Barnett and Boyce, '95). Both studies obtained subjects through support groups and not through a population-based cohort. The estimates for caregivers of children with spina bifida were calculated as differences in work time reported by spina bifida caregivers relative to the average for all workers in the southern United States. The estimates for caregivers of children with Down syndrome were calculated using cases and controls. However, data for the cases and controls were collected in different time periods: 1981–1982 for the control group and 1987–1988 for the families with a child with Down syndrome.

To convert reductions in work into caregiver time costs, an average price of time or wage rate must be assigned. Determination of the appropriate wage rate is somewhat controversial (Luce et al., '96). Wage rates differ on the basis of individual characteristics, especially sex, education, race, and age. Further, caregivers

of children with birth defects may have less time available to work and thus earn a lower wage. Other caregivers may value their time in home activities more than the wage they could earn and thus, do not work for a wage. Using individual characteristics to generate an average wage rate could result in lower time costs for some population groups.

Nevertheless, an expert panel convened by the U.S. Public Health Service to standardize cost measurements for cost-effectiveness analyses argued for wage rates based on individual characteristics (Luce et al., '96). Thus, the caregiver time costs for an individual caring for a child with a birth defect can be summarized as:

$$\text{TimeCost} = \sum_t \frac{wH'_t}{(1+r)^t}$$

where t represents the period of time that the child requires caregiver assistance in excess of the average child, H' is the difference in average hours worked per week by a caregiver of a child with a given birth defect relative to the caregiver of an average child, w is the adjusted wage rate (adjusted by age, education, and sex), and r is the discount rate. Caregiver time costs are discounted because all future costs must be valued in present terms. For example, a promise to pay \$100 2 years into the future will not be worth as much as \$100 paid presently. Requiring \$121 to be paid 2 years into the future rather than \$100 presently provides an implicit discount rate (10%). The panel on cost-effectiveness research recommends a discount rate of 3%, although many studies have used 5% (Lipscomb et al., '96).

To illustrate the calculation of caregiver time costs, we solve the formula for the estimates of work reduction described above. In both cases, we assume the reduction in work time occurs from the birth of the child through age 21. No estimates exist in the literature to guide this assumption. The study of caregivers of children with spina bifida assumed the difference would occur until age 25. We use an average wage rate of \$13 per hour (the average wage rate for private U.S. manufacturing industries) because specific wage rates for parents of children with birth defects are not known, and then we calculated costs under the assumption of a 3% and 5% discount rate.

As an example, we estimated caregiver time costs for a case of spina bifida using the assumptions described above. When averaged across all different types of lesions, this cost amounted to \$197,991 when a 3% discount rate is used. When the discount rate is changed to 5%, the cost per case decreased to \$164,675. The much smaller estimated reductions in work time for mothers of children with Down syndrome have large effects on the estimated caregiver time costs. Caregiver time costs for children with Down syndrome are estimated at \$57,203–\$68,776 depending on whether a 3%

or 5% discount rate is used. These figures should be added to the total cost of a case of spina bifida or Down syndrome.

The cost of a case of spina bifida varies depending on whether the study employs a cost-benefit or cost-effectiveness framework. In a cost-effectiveness analysis, to avoid double counting, productivity costs (lost wages for the child due to the birth defect) are captured as changes in quality adjusted life years (QALYs) in the denominator of the cost-effectiveness ratio and not in the cost calculation. In a recent *cost-effectiveness* analysis of folate supplementation, the medical, developmental, and special education costs for spina bifida (excluding productivity costs) was approximately \$150,000 per case, whereas the estimated caregiver time costs was approximately \$250,000 per case. Thus, the caregiver time costs exceeded the direct medical, developmental, and special education costs of spina bifida under a cost-effectiveness framework (Kelly et al., '96).

Under a *cost-benefit* framework, the most recent data suggest the direct and indirect costs (including productivity costs) in 1992 dollars are \$294,000 per case of spina bifida using the assumption of a 5% discount rate and \$451,000 per case of Down syndrome. Both estimates should be increased (under a societal perspective) to include the caregiver time costs, if indeed, the estimates of work reduction for these two birth defects are appropriate.

TRANSLATING FAMILY IMPACT INTO ECONOMIC VALUES

Although many consider caregiver time to be an impact on the family, distinguishing potential difficulties beyond this cost is important. Several domains of family life can be potentially affected by birth defects.

Birth defects can affect the primary caregivers of the child as well as other members of the family (Dorner, '75; Breslau et al., '82; Breslau et al., '81). Caring for a child with a disabling health condition can result in changes in the economic status of the family; limitations on the quality and quantity of interactions outside the family; conflicts and strain within the family unit, including parental and sibling relationships; and emotional distress directly related to the demands of the illness or disability. Impacts on the family differ according to the type of birth defect. For example, congenital heart defects result in more family difficulties and stress than other conditions such as craniofacial anomalies (Emery, '89).

Caring for a child with a birth defect can also affect marital quality. Marriages of parents of children with spina bifida were more conflictive when parenting stress and the strain of combining parenting with other social roles were greatest (Cappelli et al. '94). Similarly, mothers of children with craniofacial anomalies reported higher degrees of marital conflict than did mothers of other children matched for age and socioeconomic status (Speltz et al, '90). In one study, parents

of children with spina bifida reported more psychosocial stress but no differences in marital satisfaction than did parents of control children (Holmbeck et al., '97).

Some studies have indicated that although children with birth defects require substantial time and energy from the family, the amount depends greatly on the ability of the child to perform basic activities of daily living. The impact on the family of children with spina bifida is much greater for those whose children are limited in their functioning in the home (McCormick et al., '86).

The existing literature on family impact, however, will *not* aid in the development of values that can inform policy through cost-benefit or cost-effectiveness analysis. To inform cost-benefit or cost-effectiveness analysis, the impact on the family must be translated into a cost or QALY estimate. Some have suggested that if these impacts could be translated into economic values, the resulting costs might exceed traditional costs associated with birth defects (Waitzman et al., '96). Despite this acknowledgement, no research exists that documents the impact of birth defects on the family *that can be translated into a cost estimate or a QALY estimate*. Indeed, the Panel on Cost-Effectiveness in Health and Medicine summed up this issue in its cost-effectiveness analysis of folate supplementation by stating:

We have not included the psychological and physical impacts on parents and families with NTD-affected pregnancies . . . on caregivers of either children or adults with spina bifida; or on caregivers of people disabled by vitamin B12-related neurological complications. Although quality-of-life impacts on these parties are clearly important, their inclusion is not recommended because the methods for capturing these impacts are in early stages of development. Also, data on these quality-of-life impacts are not available. Including these QALYs would increase the benefits of all interventions (Kelly et al., '96).

The increasing recognition of the importance of informal caregiving has generated some research in this area. Mohide and colleagues ('88) developed an instrument to measure quality of life in caregivers—the Caregiver Quality of Life Instrument. Drummond and colleagues ('91) used this instrument to evaluate a support program for informal caregivers of demented elderly patients that found a clinically important, but not statistically significant difference in caregiver QALYs. The lack of significant findings may be attributed to the intractability of measuring caregiver difficulties for patients with dementia, or the stage of disease may have been too late for the intervention to affect caregivers.

Neumann and colleagues ('99) provided the first study to examine whether caregiver burden can be measured directly from existing generic instruments. They measured QALYs of patients suffering from Alzheimer's and of their caregivers. QALYs were measured by the Health Utilities Index Mark II (HUI:2). In

a cost-effectiveness framework, decrements (or improvements) in the QALYs of caregivers could be added to (or subtracted from) the denominator of the cost-effectiveness ratio. (Changes in caregiver time costs would be measured in the numerator). The study found significant differences in QALYs between six stages of disease (questionable, mild, moderate, severe, profound, terminal) for *patients*, but no significant differences for *caregivers*. The findings on caregivers may be attributed to the inability of the HUI:2 to detect subtle but important changes or a lack of statistical power to detect such differences (Neumann et al., '99).

Finally, many of the issues in estimating caregiver time costs and difficulties, including the possibility of double counting, are now recognized (Brouwer et al., '99). Double counting can occur if an analyst includes costs that overlap. For example, disease burden can reduce a patient's quality of life and his or her ability to earn wages. For these reasons, only QALYs are considered in standard cost-effectiveness methodology and not productivity losses. The possibility that caregiver difficulties and caregiver time costs represent a similar example of double counting is the rationale for current recommendations not to include both measures in economic analyses. In addition, because of some of the problems described in the previous section, they recommend more work on developing instruments to measure impacts directly through changes in QALYs of caregivers.

CONCLUSIONS AND RESEARCH RECOMMENDATIONS

Recent analyses of the costs of birth defects highlight the importance of cost estimates for guiding the allocation of scarce public resources to prevent birth defects. Information about the association between consumption of folic acid and neural tube defects presented policy makers with choices of whether and how folic acid consumption should be increased in women of childbearing age. Without estimates of the costs of birth defects, policy makers could not assess the benefits of other policies to increase folic acid consumption.

Cost estimates related to birth defects need to include impacts on caregivers as well. A number of issues have limited the incorporation of caregiver costs in economic analyses. Few studies have examined caregiver time costs in relation to birth defects and the most recent estimates for spina bifida are fairly old, from a limited sample, and unpublished. Yet, where these impacts have been included, the caregiver costs exceeded the traditional economic costs.

Although studies have documented increased difficulties associated with caring for children with birth defects, estimates that can be used in economic studies are lacking. Researchers in other settings, especially schizophrenia and Alzheimer's, have proposed measuring caregiver QALYs directly to estimate difficulties in economic terms for cost-effectiveness analyses. Esti-

ating these impacts using contingent valuation methods may also be possible. Contingent valuation studies are used frequently in economic evaluations and typically include measures of willingness to pay (Pauly, '96). Studies could be conducted on families of children with birth defects and control families that compare how much they would be willing to pay for informal care. Estimates of the difference could be directly incorporated into cost-benefit analyses of birth defect prevention programs.

Clearly, new evidence of the full cost of caring for children with birth defects is warranted. Developing such evidence, however, requires innovative data collection strategies and funding from either federal or state sources. Periodic surveys from the National Center for Health Statistics targeted toward children with birth defects could produce sample sizes of sufficient magnitude with national representation. Alternatively, coordinated efforts involving a number of states using consensus-based questionnaires could generate valuable data.

The birth defect prevention community has a clear incentive to invest in coordinated data collection efforts. Improved estimates of caregiver time cost and potential difficulties associated with caring for children with birth defects will assist policy makers in allocating resources for birth defect prevention and treatment.

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