Labor Market Productivity Costs for Caregivers of Children with Spina Bifida: A Population-Based Analysis

John M. Tilford, PhD, Scott D. Grosse, PhD, Allen C. Goodman, PhD, Kemeng Li, MA

Background. *Caregiver productivity costs are an important component of the overall cost of care for individuals with birth defects and developmental disabilities, yet few studies provide estimates for use in economic evaluations.* **Objective.** *This study estimates labor market productivity costs for caregivers of children and adolescents with spina bifida.* **Methods.** *Case families were recruited from a state birth defects registry in Arkansas. Primary caregivers of children with spina bifida (N = 98) reported their employment status in the past year and demographic characteristics. Controls were abstracted from the Current Population Survey covering the state of Arkansas for the same time period (N = 416). Estimates from regression analyses of labor market outcomes were used to calculate differences in hours worked per week and lifetime costs.* **Results.** *Caregivers of children with spina bifida worked an annual average of 7.5 to 11.3 hours less per week depending on the disability severity. Differences in work hours by caregivers of children with spina bifida translated into lifetime costs of $133,755 in 2002 dollars using a 3% discount rate and an age- and sex-adjusted earnings profile. Including caregivers' labor market productivity costs in prevention effectiveness estimates raises the net cost savings per averted case of spina bifida by 48% over the medical care costs alone.* **Conclusions.** *Information on labor market productivity costs for caregivers can be used to better inform economic evaluations of prevention and treatment strategies for spina bifida. Cost-effectiveness calculations that omit caregiver productivity costs substantially overstate the net costs of the intervention and underestimate societal value.* **Key words:** *lifetime costs; productivity; cost-effectiveness analysis; disability, children.* **(Med Decis Making XXXX;XX:xx–xx)**

The US Panel on Cost-Effectiveness in Health and Medicine recommended that caregiver time be valued and included in cost-effectiveness analyses conducted from the societal perspective.[1] However, although unpaid care provided by family and friends constitutes a major component of the cost of care for many chronic diseases and disabling conditions, these costs are routinely excluded from published economic evaluations.[2–4] The exclusion of caregiver time costs and other "family spillover effects" from cost-effectiveness analyses may lead analysts to understate the value of prevention.[5] Informal caregiving reduces time available for work, home production, sleep, and leisure.[6] Loss of work time due to caregiving responsibilities is important both because of its impact on family income[7] and as "a component of how a family uses *time* resources" (italics in original).[8]

Caregiver time costs are particularly relevant to economic evaluations of interventions to prevent disability in children. Although time costs include lost time in a variety of activities,[2] most analyses have been restricted to estimates of lost earnings. Anderson and others provide a recent literature

Address correspondence to John M. Tilford, PhD, Center for Applied Research and Evaluation, 800 Marshall St., Little Rock, AR 72202-3591; e-mail: tilfordmickj@uams.edu.

review on the personal costs to families of caring for a child with physical disabilities such as spina bifida and cerebral palsy,[8] which addresses equity issues associated with caring for disabled children, especially the question of whether such families experience a disproportionate burden. They found few studies of the magnitude of costs imposed on these families. There was considerable variability in costs due to incomplete reporting of the severity of the disability or disability categories.

Similar issues arise in the literature on child disability and maternal labor market outcomes. Some studies find a large impact of child disability on maternal labor market outcomes,[9−11] others a more modest effect.[12] No published study estimates the lifetime productivity costs associated with caring for a child with spina bifida.

The purpose of this study is to provide an estimate of lifetime labor market productivity costs for caregivers of children with spina bifida to be used in cost-effectiveness evaluations of interventions such as folic acid fortification of foods or promotion of preconception vitamin supplement use.[13,14] Spina bifida is a disabling condition of childhood that results from the incomplete closure in utero of the tissue and bone surrounding the spinal cord. It is associated with an elevated risk of death, especially in infancy, and with multiple surgeries. Children born with spina bifida can have mild to severe disabilities depending on the location of the lesion. Children with lower or sacral malformations may only have bowel and bladder dysfunction, whereas higher lumbar or thoracic lesions can cause varying degrees of limb paralysis among other disabilities. Spina bifida and anencephaly are related neural tube defects (NTDs) whose risk is reduced by adequate intake of folic acid prior to pregnancy.[15−17] Since 1998, both the United States and Canada have mandated the fortification of certain foods with folic acid to reduce the incidence of NTDs. Evidence to date suggests significant reductions in the number of births affected by spina bifida following fortification.[13] Similar policies have not yet been adopted in most other industrialized countries.

One (unpublished) study of labor market productivity costs for caregivers of children with spina bifida has been reported previously[18] and used in a cost-effectiveness evaluation comparing strategies to prevent neural tube defects.[14] Its findings were based on labor market outcomes for families residing in North Carolina that participated in spina bifida support groups. The use of support groups to obtain subjects might have inflated estimates of labor market productivity costs, because membership in a support group could reflect more leisure time available from reduced work time.[19]

METHODS

Model

This study values labor market productivity losses using hours of paid employment foregone, multiplied by the real market hourly wage rate, including the value of fringe benefits. Our objective is to develop estimates of labor supply decisions for caregivers of children with spina bifida relative to caregivers of children from the population. Lifetime or incidence-based costs are preferred in prevention effectiveness models.[20] Miller and others[21] define *incidence-based costs* as the present value of the lifetime costs that may result from injuries that occur during a single year. They argue that incidence-based costs measure the savings that successful prevention efforts can yield.

Several prevention effectiveness studies have reported costs of medical care or labor market productivity costs using a lifetime approach.[21−24] Lifetime models typically use cross-sectional data from a prevalent population to construct an age-specific profile of costs rather than attempting to follow an incident cohort over a long period of time.[20,25] Caregiver productivity costs associated with children older than 1 year require discounting to generate estimates in present values. Resulting estimates yield a lifetime cost per case averted that can be used to assess the prevention effectiveness of health interventions.

A simple conceptual model comparing lifetime productivity costs (LPC) for caregivers of children with spina bifida to other caregivers is illustrated by the following:

$$\text{LPC} = \sum_t \frac{w_t(s_t^{sb}H_t^{sb} - s_t^{o}H_t^{o})}{(1+r)^t}. \qquad (1)$$

In this equation, parameter s_t is a survival probability for the child; w_t is the opportunity cost of labor, defined as the age- and gender-adjusted hourly real wage that could be earned in the absence of caring for a disabled child including fringe benefits; and H_t is the average hours worked over the year. Superscript sb refers to caregivers of children with spina bifida, and superscript o refers to caregivers of children from the population. Variable r is the discount rate (here, 3%), and t is the age of the child defined

over the relative age range in which the child's disability is likely to impact labor market outcomes.[26]

Our primary interest in this study is obtaining statistical estimates of the difference in average hours worked for caregivers of spina bifida relative to caregivers from the population. Calculation of average hours worked in a statistical model is achieved through 3 equations. Average hours worked per year can be influenced by the caregiver's likelihood of employment (p), the number of hours worked per week (N), and the number of weeks worked in a given year (V), assuming employment E. If $E^* > 0$ is a latent variable, then

$$H_t^{sb} = [(p(\hat{E})|D=1)x(\hat{V}|D=1, E^*>0)x(\hat{N}|D=1, E^*>0)],$$
(2a)

and similarly for the general population,

$$H_t^o = [(p(\hat{E})|D=0)x(\hat{V}|D=0, E^*>0)x(\hat{N}|D=0, E^*>0)],$$
(2b)

where D symbolizes an indicator variable with $D=1$ for caregivers of children with spina bifida (cases) and $D=0$ for caregivers from the population (controls). Thus, the difference in hours worked averaged over the year (not average hours worked per week conditional on working as described below) between cases and controls can be defined as

$$\Delta H_t = H_t^{sb} - H_t^o.$$
(2c)

In practice, fitted values for caregiver employment probabilities ($\hat{p}$), average hours worked per week ($\hat{N}$), and weeks worked per year ($\hat{V}$) are used to calculate the estimated values for Equations 2a, 2b, and 2c.

Estimation of $\hat{p}$ assumes an underlying response variable (E^*) defined by the discrete choice (typically either probit or logit) regression relationship

$$E^* = \beta x + \alpha D + \mu.$$
(3)

Thus, we observe whether the caregiver is employed according to

$$E = 1 \text{ if } E^* > 0$$

and

$$E = 0 \text{ otherwise.}$$

In addition, we estimate the number of weeks worked (V) and the hours worked per week (N)

conditional on $E^* > 0$ and the least squares regression relationships

$$v = \beta X + \alpha D + \varepsilon \text{ if } E^* > 0,$$
(4)

and

$$N = \delta X + \gamma D + \eta \text{ if } E^* > 0.$$
(5)

As noted above, in Equations 3, 4, and 5, the variable set x refers to labor market determinants common to both case and control caregivers, with D defining an indicator variable to describe cases and controls.

The conceptual model implies that the impact on hours worked comes from a single type of spina bifida, but, in reality, the impact can vary by type and severity. We generate estimates for 3 levels of severity consistent with prior work: sacral lesions, lower lumbar lesions, and upper lumbar/thoracic lesions with all the children having a single type of spina bifida (aperta). The resulting estimates of prevalent costs by child age and severity form the basis for a simulation model of lifetime costs consistent with equation 1.

Our empirical analysis relies on 3 regressions. We use a logit regression to estimate the probability of employment and ordinary least squares (OLS) regression to estimate the number of weeks worked and the number of hours worked per week. Following Bradley and others,[27,28] we do not attempt to address selection issues with this database because of the lack of variables that could be omitted a priori from the 2nd stage of a 2-stage model. We also do not estimate wage equations because of the limited frequency of responses to this question in the study samples.

Calculation of ΔH_t, or the difference in hours worked between caregivers of children with spina bifida and population controls over the t years when disability can influence labor market outcomes, in the absence of expensive longitudinal cohort data, requires that data on a prevalent population be transformed for analysis as incidence-based estimates.[20] We accomplished this transformation by using predicted values from equations 3 to 5 to obtain estimated differentials in average hours worked (averaged over the year, not the conditional average for working caregivers) for each of the case caregivers relative to the average for the control caregivers. We then estimated the relationship between age of the affected child and the difference in average hours worked for all caregivers of children with spina bifida. The resulting profile captures differences in hours worked

from birth to age 21 (with out-of-sample predictions) that can be used to simulate lifetime costs. This profile is the same as if an incident cohort had been followed over a 21-year period; it provides an estimate of the labor market productivity costs avoided by preventing a neural tube–affected birth.

Data

The study utilizes a case-control design with case families developed from a unique sample of families residing in the state of Arkansas that were part of a population-based registry of birth defects in operation for over 20 years. The Arkansas Reproductive Health Monitoring System (ARHMS) is a unique birth defect registry containing population-based information for children up to 20 years of age. We used AHRMS to identify all families caring for a child with spina bifida aged 0 to 17 who resided in Arkansas. A letter announcing plans for the study was sent to the family followed by a telephone call asking for consent to be interviewed. All data pertaining to case families were obtained by telephone interviews with the primary caregiver over a 2-year period, 2001 to 2002. The interviewer asked questions about the health status of both the child with spina bifida and the primary caregiver in addition to the employment status of the primary caregiver. A detailed description of the study design and interview response rate has been published elsewhere.[29] Among identified families with valid phone numbers, the response rate was 82%. In the larger sample, including families where a valid phone number could not be obtained, the response rate was 47%.

In the section of the interview on employment status, caregivers were asked to report whether they were employed in the past year, how many weeks they worked in the past year (weeks of paid vacation counted as time worked so that a caregiver working a full year would have worked 52 weeks), and how many hours per week they worked, on average, if they did work. The wording of the questions was identical to recent national health surveys and follows closely the wording in the Current Population Survey (CPS). In addition, questions were asked about the location of the lesion for affected children and demographic information.

The sampling strategy allows the data obtained from the case families to be compared with data collected as part of the CPS. Because of this comparability, control families for the study could be abstracted from respondents to the CPS from the state of Arkansas for the years in which the interviews took place. Data on households with either a married woman or female head with at least 1 child present under the age of 18 years were selected from the Arkansas portion of the March supplement to the CPS for 2001 to 2002. Because more case families were interviewed in 2002 than 2001, data were randomly dropped from the CPS for 2001 to ensure that the proportions of interviews conducted in each year were similar in both samples. This sampling strategy resulted in 98 cases obtained from the Arkansas birth defect registry (41 with sacral lesions, 33 with lower lumbar lesions, and 24 with upper lumbar/thoracic lesions) and 416 controls from the CPS. All regression equations used the form:

$$y = a + \sum_i b_i X_i + c_1 D_1 + c_2 D_2 + c_3 D_3, \qquad (6)$$

with D_1 referring to sacral lesions, D_2 to lower lumbar lesions, and D_3 referring to upper lumbar/thoracic lesions.

The CPS is an excellent source of controls for studies of labor market outcomes because it is population based and has a large number of observations available for any particular state or region. Bradley and others[30] recently described an agenda for research on labor market outcomes of cancer survivors that can be applied to other chronic conditions based on using the CPS as a source of controls for case data from population-based registries. Our research design closely follows this agenda.

We conducted regression analyses using both case and control families while controlling for location of the lesion through dummy variables. Because disease severity for spina bifida increases with the height of the lesion on the child's back, we expected an increase in productivity costs for a child with higher level lesions. In addition to lesion location, controls for caregiver age, age squared, the number of children under 5 years of age, the number of children aged 5 to 18 years, education, marital status, and race were included in the 3 regressions estimating probability of working, weeks worked per year, and hours worked per week.

A primary objective of the analysis is to generate caregiver productivity cost estimates that are comparable with recent estimates of the medical care costs associated with spina bifida by Waitzman and others.[31] Their analysis relied on relative survival probabilities for children[22] with spina bifida that have recently been updated[32] as survival in this population has improved over time. Thus, we first estimate caregiver productivity costs in 2002 dollars

Table 1 Characteristics of Caregivers and Their Children

Characteristic	Case Caregivers	Control-CPS Caregivers
Caregiver age	37.7 (8.8)	35.3 (8.1)[a]
Caregiver race		
Black (%)	6.1	18.0[a]
White (%)	90.8	81.0[b]
Other (%)	3.1	1.0
Caregiver education		
<High school education (%)	11.2	12.5
High school graduate (%)	41.8	41.1
College or trade (%)	29.6	25.5
College graduate (%)	17.4	20.9
Caregiver marital status		
Divorced (%)	13.3	9.6
Married (%)	77.5	75.0
Other (%)	9.2	15.4
Number of children		
<5 years of age	0.61 (0.93)	0.49 (0.73)
5–18 years of age	1.64 (1.04)	1.53 (1.07)
Child <5 years present (%)	39.8	37.3
Lesion location		
Sacral	41.8	—
Lower lumbar	33.7	—
Upper lumbar/thoracic	24.5	—
Labor market outcomes		
Wage rate	10.5	13.9[b]
Worked in past year (%)	54.1	76.7[a]
Weeks worked past year[c]	40.4 (16.5)	44.8 (13.5)[b]
Hours worked per week[c]	36.0 (9.7)	37.0 (9.3)
N	98	416

Note: CPS = Current Population Survey. Values are presented as mean (SD).
a. Significant at 0.01 level.
b. Significant at 0.05 level.
c. Conditional on having worked in past year.

using the same set of relative survival probabilities for children with spina bifida as the Waitzman analysis to make our productivity cost estimates comparable with their direct cost estimates. Then we use new relative survival probabilities and adjust to 2005 dollars to provide up-to-date estimates for cost-effectiveness analysis.

In addition to survival probabilities, calculation of lifetime caregiver productivity cost requires data on the age-earnings profile of caregivers. As with a previous analysis, we used an age-earnings profile in which, consistent with the data, the average caregiver at birth is a 25-year-old female who ages to 46 years after 21 years.[14] We rely on data on average female earnings in 2000 by age for *all* workers including fringe benefits, to reflect the marginal social product of labor, with an assumed 1% per year increase in real labor productivity.[33] We adjust these values to 2002 and 2005 dollars using the Employment Cost Index.

RESULTS

Table 1 compares the characteristics of case and control families. Across most characteristics, there was little difference between the 2 groups. Caregivers were slightly older for cases and the percentage of blacks was higher among controls. The underrepresentation of blacks among case families is consistent with prevalence estimates of spina bifida in other studies.[34] Case and control families did not differ with respect to education, marital status, or number of children.

Table 1 also provides summary statistics for labor market outcomes. Without adjusting for differences in sample characteristics, case caregivers earned significantly lower wages, were much less likely to have worked in the past year, and worked fewer weeks conditional on having worked. There was no difference in the average number of hours worked

Table 2 Multivariate Regressions of Labor Market Outcomes

Characteristic	(a) Logistic *Worked Last Year*		(b) OLS *Weeks Worked*		(c) OLS *Hours Worked*	
	Marginal Δ[a]	SE	Beta[c]	SE	Beta[c]	SE
Age	0.009	(0.158)	2.045[d]	(0.601)	1.204[d]	(0.430)
Age squared	−0.0002	(0.0002)	−0.023[d]	(0.008)	−0.016[d]	(0.006)
White	−0.047	(0.053)	1.251	(1.775)	−0.695	(1.271)
Divorced	0.117[f]	(0.055)	0.916	(2.157)	3.359[e]	(1.545)
Children <5 years	−0.048	(0.031)	−2.757[e]	(1.103)	−1.798[e]	(0.790)
Children 5–18 years	0.003	(0.021)	−1.090	(0.765)	−0.913[f]	(0.547)
Not HS graduate	−0.299[e]	(0.075)	−0.056	(2.656)	1.234	(1.901)
Some college	0.010	(0.051)	2.720	(1.669)	−0.088	(1.193)
College graduate	0.029	(0.055)	3.056[f]	(1.832)	0.892	(1.312)
Sacral lesion	−0.215[d]	(0.085)	−1.872	(2.838)	−1.150	(2.033)
LL lesion	-0.255[d]	(0.094)	−6.134[f]	(3.265)	−0.085	(2.338)
HT lesion	−0.273[d]	(0.111)	−8.015[e]	(4.048)	0.314	(2.784)
Constant	—	—	2.522	(10.948)	18.038	(7.841)
R^2	0.088[b]		0.124		0.029	
N	514		372		372	

Note: OLS = ordinary least squares; SE = standard error; HS = high school; LL = lower lumbar; HT = upper lumbar/thoracic.
a. Marginal changes in probability from logistic regression evaluated at the means of the variables.
b. Pseudo R^2.
c. Estimate from linear regression.
d. Significant at the 0.01 level.
e. Significant at the 0.05 level.
f. Significant at the 0.10 level.

per week between cases and controls conditional on having worked in the past year.

Table 2 presents the findings from the multivariate regression models that predict (a) whether the caregiver worked in the prior year; (b) the number of weeks worked conditional on having worked; and (c) the hours worked per week conditional on having worked. The data from the CPS ($N = 416$) and the population-based registry ($N = 98$) were combined. Column (a) provides estimates from the logistic regression and, as expected, indicates significant impacts of having less than a high school education, the number of children under 5 years of age, and divorce on the probability of having worked in the past year. Most important, the data indicate large impacts of the 3 lesion locations on the probability of working relative to controls. Presence in the household of a child with spina bifida decreased the average probability of having worked in the last year by 21.5 (sacral lesion) to 27.3 (upper lumbar/thoracic lesions) percentage points.

Column (b) provides OLS estimates of the number of weeks worked conditional on having worked in the past year. Caregivers of children with sacral lesions did not have significantly different estimates of weeks worked relative to controls, with the point

estimate indicating less than 2 weeks' difference. In contrast, both caregivers of children with lower lumbar lesions and higher lumbar/thoracic lesions worked significantly fewer weeks relative to controls, with point estimates of 6 and 8 weeks.

Finally, column (c) provides OLS estimates of average hours worked per week conditional on having worked in the past year. As would be expected, caregiver age, the number of children under 5 years of age, and divorce were significant predictors in this model. Caregivers of children with spina bifida and controls worked similar numbers of hours per week, conditional on working, irrespective of lesion location.

In columns (b) and (c), the number of case subjects is reduced from 98 to 54 subjects. The coefficients for sacral lesion, lower lumbar lesion (LL), and upper lumbar/thoracic lesion (HT) may be less stable due to the reduction in degrees of freedom for estimation, but there is no reason to believe that they are biased or inconsistent.

Figure 1 provides the estimated relationship (and 95% confidence intervals) between age of the child and paid hours of work using a quadratic function with out-of-sample predictions for children under 2 years of age and children over 17 years of age. The confidence intervals are based on the standard errors from
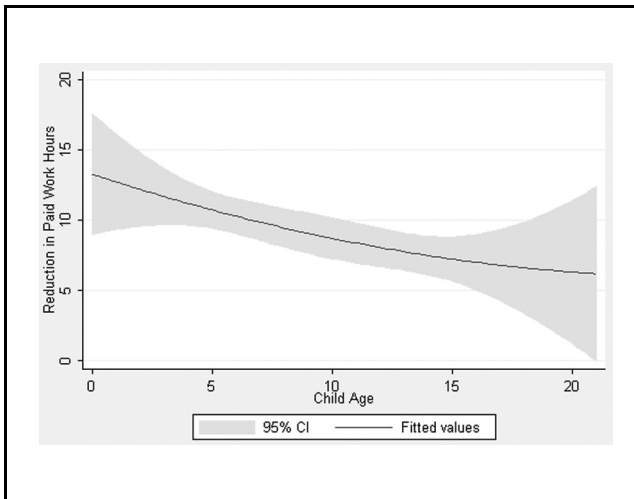
Figure 1 Reduction in paid work hours per week for caregivers of children with spina bifida relative to controls by age of affected child.

the predicted model, with the exception of the out-of-sample predictions, in which the larger forecast standard errors are substituted. The estimated relationship indicates that the reduction in paid work hours is greatest for the caregivers of the youngest children with spina bifida and lowest for the caregivers of the oldest children. The predicted reduction in paid hours worked per week (averaged over the year) ranges from 13.3 (95% confidence interval [CI], 9.1–17.5) for infants in the 1st year of life to 6.2 (95% CI, 1.3–11.3) for children 21 years of age.

Table 3 provides estimates of overall reductions in paid hours worked (per week) and lifetime foregone wages, by lesion location, using equation 1 in the text and the resulting estimates from Table 2 and the relationship in Figure 1. Column (a) provides estimated reductions in paid work for caregivers of children with spina bifida. As hypothesized, reductions in paid work were greatest for the upper lumbar/thoracic lesions (–11.3 hours) and smallest (–7.5 hours) for caregivers of children with sacral lesions. Most of the reduction results from the lower probability of being employed, with a small share due to fewer weeks worked among those in the labor market. This is consistent with previous studies of maternal labor market outcomes and child disability.[10]

Column (b) in Table 3 provides lifetime productivity costs using the predicted differences in child age obtained from Figure 1 for all children with spina bifida, and by lesion location. These estimates are in 2002 US dollars to be comparable with the

estimates of direct medical care cost for children with spina bifida by Waitzman and others.[31] Inserting the estimated differences into equation 1 results in lifetime productivity costs of $133,755 over all lesions (95% CI, $106,910–$171,184). Column (c) in Table 3 provides productivity cost estimates in 2005 US dollars based on the updated survival probabilities.[36] The updated estimate is $158,771 per case of spina bifida (95% CI, $122,412–$194,975) with most of the difference attributed to the change in wage rates between 2002 and 2005. The change in survival probabilities contributed 6.5% to the difference in cost estimates.

DISCUSSION

Estimates of the lifetime productivity costs associated with informal caregiving are important to account fully for the economic benefits of preventing childhood disability. In early work, Salkever noted the large cost of disabled children on mothers' labor supply decisions.[11] He called for "more detailed analysis of specific categories of chronic diseases" and research on the mental and physical health of parents of disabled children. Our study addresses spina bifida, a potentially disabling birth defect that can be prevented with adequate maternal intake of folic acid prior to conception.

We generated population-based samples for caregivers of children with spina bifida in Arkansas, and a control sample, by combining registry data with secondary data from the CPS following the example of Bradley and others.[30] We found a substantial impact of caring for a child with spina bifida on labor force participation, with participation rates 21% to 27% lower than those in the control group. The estimates of the difference are at the high end of the range of effects previously reported for caregivers of children with disabilities. For example, 1 study found that caregivers of children with disabilities had an adjusted marginal difference in employment probability of –6% to –11%, depending on the definition of child disability.[10] A study from Ontario, Canada, reported that caregivers of children with cerebral palsy had a 15% lower labor force participation rate than a representative sample of parental caregivers in the same province.[35] An analysis of data from the US National Health Interview Survey 1994 Disability Supplement revealed that maternal caregivers of children with an activity limitation had a 25% lower probability of employment.[36]

Table 3 Average Reduction in Hours Worked Per Week and Lifetime Labor Market Productivity Costs for Caregivers of Children with Spina Bifida[a]

Lesion Location	(a) Mean Reduction in Work Hours per Week (95% CI)	(b) Lifetime Productivity Costs (2002 US Dollars)			(c) Lifetime Productivity Costs (2005 US Dollars)[b]		
		Mean	Lower Limit	Upper Limit	Mean	Lower Limit	Upper Limit
Overall	−9.2 (−10.2, −8.2)	$133,755	$106,910	$171,184	$158,771	$122,412	$194,975
By lesion							
Sacral lesion	−7.5 (−9.1, −5.9)	$113,451	$81,243	$145,527	$129,586	$93,227	$165,790
LL lesion	−9.8 (−11.2, −8.3)	$148,164	$115,957	$180,240	$169,072	$132,713	$205,275
UL/thoracic	−11.3 (−13.4, −9.2)	$170,803	$138,596	$202,879	$194,823	$158,464	$231,027

Note: CI = confidence interval; LL = lower lumbar; UL = upper lumbar. 95% confidence interval in parentheses.
a. Lifetime estimates based on children from birth to 21 years of age.
b. Uses updated estimates of survival probabilities for children with spina bifida.

An important advantage of this study is that it provides data on caregiver productivity costs classified by the location of the child's lesion, an objective indicator of physical impairment. This variable allows for internal validation of the impact of child disability on maternal labor force outcomes that is independent of the selection of a control sample. Findings by lesion location may have added importance if prevention strategies have a disproportionate effect on the more severe lesions. Even the estimates for the least severe lesions indicate a substantial decrease in work hours and an increase in lifetime productivity costs for the primary caregiver.

The total lifetime cost of medical, developmental, and special education costs for spina bifida were previously estimated to be $279,210, in 2002 dollars.[13,31] Our estimate of caregiver labor market productivity costs during childhood of $133,755 in 2002 dollars raises the total estimate of lifetime costs to $412,965, a figure that is 48% higher. Our estimates are conservative because we lack data on productivity costs associated with caring for adults with spina bifida and because we used outdated estimates of survival to be consistent with the previous spina bifida cost study.[31] Using more recent survival estimates would raise this estimate by 6%.

Accurately accounting for family effects in cost-effectiveness analyses can improve our understanding of the full costs and benefits of prevention and treatment interventions. Although we find that caregiver labor market productivity costs constitute a major component of the lifetime costs of spina bifida, our estimate does not include other important time considerations including household production, sleep, and leisure. Inclusion of these costs increased the total cost of caregiving in a study of adults with cystic fibrosis.[37] New data sets with detailed time diary information from the representative population are now available.[38] Studies seeking to incorporate the full cost of informal caregiving into economic evaluations and other dimensions of time use and health may now be feasible with this new source of population-based data on time use.[39]

An important limitation of the study is that it uses only labor market data from the state of Arkansas. Productivity costs based on a registry from a single state may not generalize to the nation as a whole or to other regions of the country. However, the reported findings from this study, although smaller in magnitude, are broadly similar to unpublished findings from the state of North Carolina that relied on a convenience sample of caregivers participating in support groups.[18] The similarity suggests the findings may be applicable to other states and regions of the country. It should also be noted that concentration on a single state reduces unmeasured heterogeneity of labor markets, health care systems, and state policies that may confound more geographically dispersed studies.

The study also is limited in that wage data at the individual level were incomplete and did not permit the inclusion of the wage rate in modeling labor market outcomes.[40] To generate productivity cost estimates consistent with recommendations from the US Panel on Cost-Effectiveness,[14] we used age- and gender-specific estimates from previously published estimates. Such a strategy has been employed in other studies as well.[41]

Finally, the study is limited by the small sample of case families obtained from the birth defects registry. Obtaining cases from a different geographical region with a larger population could produce samples to obtain estimates with greater precision.

CONCLUSIONS

Economic analyses of fortification of foods with folic acid consistently demonstrate positive net benefits.[13] Previous analyses of the benefits of fortification contributed to mandates to fortify enriched cereal-grain grain products in the United States and Canada.[14,42] Subsequently, the birth prevalence of spina bifida dropped in both countries by over 30%, a much greater decline than had been projected in economic analyses published prior to fortification.[13] The postfortification economic analysis calculated likely net cost savings in 2002 dollars of $143 million per year as a result of averted births with spina bifida or anencephaly. Adding our conservative estimate of caregiver productivity cost raises the estimated annual cost savings from fortification to $228 million. Despite economic evaluations that show the benefits of fortifying food with folic acid greatly exceed the costs, food fortification still has not been adopted in most industrialized countries.[43,44] The inclusion of caregiver productivity costs and other caregiver impacts[29] could provide additional impetus for folic acid fortification in countries that have not adopted such a policy.

The inclusion of caregiver productivity costs is more likely to alter cost-effectiveness ratios for other prevention strategies, notably promotion of preconception folic acid or multivitamin intake, that are less likely to be cost saving than fortification. Cost-effectiveness calculations underestimate their societal value if caregiver time costs and other potential spillover effects are not included. For example, a targeted approach to prevent recurrent neural tube defects by identifying women with a prior affected pregnancy and counseling them to consume high-dose folic acid supplements has been shown to be effective,[45] but the cost effectiveness of this labor-intensive approach remains to be demonstrated. This study's findings can be used to incorporate estimates of caregiver labor market productivity costs in such an evaluation. Additional research is needed on other components of caregiver time use such as sleep, home production, and leisure to capture the full time cost of caring for children with spina bifida.

REFERENCES

1. Luce BR, Manning WG, Siegel JE, Lipscomb J. Estimating costs in cost-effectiveness analysis. In: Gold MR, Siegel JE, Russell LB, Weinstein MC, eds. Cost-Effectiveness in Health and Medicine. New York, Oxford: Oxford University Press; 1996. p 176–213.

2. van den Berg B, Brouwer WB, Koopmanschap MA. Economic valuation of informal care. An overview of methods and applications. Eur J Health Econ. 2004;5:36–45.

3. Stone PW, Chapman RH, Sandberg EA, Liljas B, Neumann PJ. Measuring costs in cost-utility analyses. Variations in the literature. Int J Technol Assess Health Care. 2000;16:111–24.

4. Tranmer JE, Guerriere DN, Ungar WJ, Coyte PC. Valuing patient and caregiver time: a review of the literature. Pharmacoeconomics. 2005;23:449–59.

5. Basu A, Meltzer D. Implications of spillover effects within the family for medical cost-effectiveness analysis. J Health Econ. 2005;24:751–73.

6. Brouwer WBF, Van Exel NJA, Koopmanschap MA, Rutten FFH. The valuation of informal care in economic appraisal. Int J Technol Assess Health Care. 1999;15:147–60.

7. Breslau N, Salkever D. Women's labor force activity and responsibilities for disabled dependents: a study of families with disabled children. J Health Soc Behav. 1982;23:169–83.

8. Anderson D, Dumont S, Jacobs P, Azzaria L. The personal costs of caring for a child with a disability: a review of the literature. Public Health Rep. 2007;122:3–16.

9. Gould E. Decomposing the effects of children's health on mother's labor supply: is it time or money? Health Econ. 2004;13:525–41.

10. Powers ET. Children's health and maternal work activity: estimates under alternative disability definitions. Journal of Human Resources. 2003;38:522–56.

11. Salkever DS. Children's health problems: implications for parental labor supply and earnings. In: Fuchs VR, ed. Economic Aspects of Health. Chicago: University of Chicago Press; 1982. p 221–51.

12. Carmichael F, Charles S. The labour market costs of community care. J Health Econ. 1998;17:747–65.

13. Grosse SD, Waitzman NJ, Romano PS, Mulinare J. Reevaluating the benefits of folic acid fortification in the United States: economic analysis, regulation, and public health. Am J Public Health. 2005;95:1917–22.

14. Kelly AE, Haddix AC, ScanlonKS, Helmick CG, Mulinare J. Cost-effectiveness of strategies to prevent neural tube defects. In: Siegel JE, Russell LB, Weinstein MC, Gold MR, eds. Cost-Effectiveness in Health and Medicine. New York, Oxford: Oxford University Press; 1996. p 312–49.

15. Prevention of neural tube defects: results of the Medical Research Council Vitamin Study. MRC Vitamin Study Research Group. Lancet. 1991;338:131–7.

16. Berry RJ, Li Z, Erickson JD, et al. Prevention of neural-tube defects with folic acid in China: China-U.S. Collaborative Project for Neural Tube Defect Prevention [corrected; erratum to be published]. N Engl J Med. 1999;341:1485–90.

17. Czeizel AE. Primary prevention of neural-tube defects and some other major congenital abnormalities: recommendations for the appropriate use of folic acid during pregnancy. Paediatr Drugs. 2000;2:437–49.

18. Lipscomb J. Human capital, willingness-to-pay, and cost-effectiveness analyses of screening for birth defects in North Carolina. Presented at the annual meeting of the American Economic Association, December 1986, New Orleans.

19. Tilford JM, Robbins JM, Hobbs CA. Improving estimates of caregiver time cost and family impact associated with birth defects. Teratology. 2001;64(Suppl 1):S37–41.

20. Haddix AC, Teutsch SM, Phaedra S. Corso PS, eds. Prevention Effectiveness: A Guide to Decision Analysis and Economic Evaluation. New York: Oxford University Press; 1996.

21. Miller TR, Romano EO, Spicer RS. The cost of childhood unintentional injuries and the value of prevention. Future Child. 2000;10:137–63.

22. Waitzman NJ, Scheffler RM, Romano PS. The Costs of Birth Defects: Estimates of the Value of Prevention. New York, London, Lanham: University Press of America Inc.; 1996.

23. Berkowitz N, Gupta S, Silberman G. Estimates of the lifetime direct costs of treatment for metastatic breast cancer. Value Health. 2000;3:23–30.

24. Chen L, Hay JW. Cost-effectiveness of primary implanted cardioverter defibrillator for sudden death prevention in congestive heart failure. Cardiovasc Drugs Ther. 2004;18:161–70.

25. Alemayehu B, Warner KE. The lifetime distribution of health care costs. Health Serv Res. 2004;39:627–42.

26. Lipscomb J, Weinstein MC, Torrance GW. Time preference. In: Gold MR, Siegel JE, Russell LB, Weinstein MC, eds. Cost Effectiveness in Health and Medicine. New York, Oxford: Oxford University Press; 1996. p 214–46.

27. Bradley CJ, Neumark D, Bednarek HL, Schenk M. Short-term effects of breast cancer on labor market attachment: results from a longitudinal study. J Health Econ. 2005;24:137–60.

28. Bradley CJ, Bednarek HL, Neumark D. Breast cancer survival, work, and earnings. J Health Econ. 2002;21:757–79.

29. Tilford JM, Grosse SD, Robbins JM, Pyne JM, Cleves MA, Hobbs CA. Health state preference scores of children with spina bifida and their caregivers. Qual Life Res. 2005;14:1087–98.

30. Bradley CJ, Neumark D, Oberst K, Luo Z, Brennan S, Schenk M. Combining registry, primary, and secondary data sources to identify the impact of cancer on labor market outcomes. Med Decis Making. 2005;25:534–47.

31. Waitzman NJ, Romano PS, Grosse SD. The half-life of cost of illness estimates: the case of spina bifida. In: Wyszynski DF, ed. Neural Tube Defects: From Origin to Treatment. New York: Oxford University Press; 2005. p 342–51.

32. Wong LY, Paulozzi LJ. Survival of infants with spina bifida: a population study, 1979–94. Paediatr Perinat Epidemiol. 2001;15: 374–8.

33. Grosse SD. Productivity loss tables. In: Haddix AC, Teutsch SM, Corso PS, eds. Prevention Effectiveness: A Guide to Decision Analysis and Economic Evaluation. 2nd ed. London: Oxford University Press; 2003. p 245–57.

34. Williams LJ, Rasmussen SA, Flores A, Kirby RS, Edmonds LD. Decline in the prevalence of spina bifida and anencephaly by race/ethnicity: 1995–2002. Pediatrics. 2005;116:580–6.

35. Brehaut JC, Kohen DE, Raina P, et al. The health of primary caregivers of children with cerebral palsy: how does it compare with that of other Canadian caregivers? Pediatrics. 2004;114: e182–91.

36. Kuhlthau KA, Perrin JM. Child health status and parental employment. Arch Pediatr Adolesc Med. 2001;155:1346–50.

37. Guerriere DN, Tullis E, Ungar WJ, et al. Economic burden of ambulatory and home-based care for adults with cystic fibrosis. Treat Respir Med. 2006;5:351–9.

38. Hamermesh DS, Frazis H, Stewart J. The American time use survey. J Econ Perspect. 2005;19:221–32.

39. Russell LB, Ibuka Y, Abraham KG. Health-related activities in the American Time Use Survey. Med Care. 2007;45:680–5.

40. Ettner SL. The impact of "parent care" on female labor supply decisions. Demography. 1995;32:63–80.

41. Cantor SB, Levy LB, Cardenas-Turanzas M, et al. Collecting direct non-health care and time cost data: application to screening and diagnosis of cervical cancer. Med Decis Making. 2006;26: 265–72.

42. Romano PS, Waitzman NJ, Scheffler RM, Pi RD. Folic acid fortification of grain: an economic analysis. Am J Public Health. 1995;85:667–76.

43. Busby A, Armstrong B, Dolk H, et al. Preventing neural tube defects in Europe: a missed opportunity. Reprod Toxicol. 2005; 20:393–402.

44. Oakley GP, Mandel JS. Folic acid fortification remains an urgent health priority. BMJ. 2004;329:1376.

45. Stevenson RE, Allen WP, Pai GS, et al. Decline in prevalence of neural tube defects in a high-risk region of the United States. Pediatrics. 2000;106:677–83