

# Health Care Expenditures of Children and Adults with Spina Bifida in a Privately Insured U.S. Population

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**BACKGROUND:** We provide new estimates of medical care utilization and expenditures over the lifespan for persons living with spina bifida in the United States. Updated estimates are essential for calculations of lifetime costs and for economic evaluations of prevention and management strategies for spina bifida. **METHODS:** We analyzed data from the 2001–2003 MarketScan database on paid medical and prescription drug claims of persons covered by employer-sponsored health insurance in the United States. Medical care utilization and expenditures during 2003 were analyzed for persons with a diagnosis of spina bifida recorded during 2001–2003 who had 12 months of coverage in a fee-for-service health plan. To calculate expenditures during infancy, a separate analysis was performed for those born during 2002 with claims and expenditures data during the first 12 months of life. We compared medical expenditures for persons with and without spina bifida by age groups. **RESULTS:** Average incremental medical expenditures comparing patients with spina bifida and those without were \$41,460 per year at age 0, \$14,070 at ages 1–17, \$13,339 at ages 18–44, and \$10,134 at ages 45–64. Children ages 1–17 years with spina bifida had average medical expenditures 13 times greater than children without spina bifida. Adults with spina bifida had average medical expenditures three to six times greater than adults without spina bifida in this privately insured population. **CONCLUSIONS:** Although per capita medical care utilization and expenditures are highest among children, adults constitute an important and growing share of the population living with spina bifida. *Birth Defects Research (Part A) 79:552–558, 2007.* © 2007 Wiley-Liss, Inc.<sup>†</sup>

**Key words:** spina bifida; medical care utilization; incremental medical expenditures; lifetime costs; economic evaluation

## INTRODUCTION

Spina bifida is a congenital malformation of the spinal cord that results from failure of fusion of the caudal neural tube during the first 2 weeks of embryonic development. Although spina bifida is a birth defect, it is not strictly a pediatric condition. It is a complex and often disabling condition that requires frequent medical attention throughout the lifespan. Persons receiving medical care for spina bifida are increasingly likely to be adults. The change in demographic composition reflects in part decreases in the number of children born with spina bifida in the United States as a result of improved diets, as well as increased use of prenatal diagnosis (Yen et al., 1992). In recent years, increased intakes of folic acid from supplements and fortified foods have contributed to the decline (Canfield et al., 2005; Williams et al., 2005). Of equal importance, children born with spina bifida are more likely to survive to adulthood as the result of

advances in medical technology and practice (Wong et al., 2001; Bol et al., 2006).

With a growing number of adults living with spina bifida and changing patterns of treatment, more accurate assessment is needed of the current expenditures on medical services for this population throughout their life-

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spans. This information is needed for the calculation of the lifetime cost of spina bifida, which is essential for economic evaluations, such as cost-effectiveness of interventions to prevent spina bifida. Previous estimates of the lifetime cost of spina bifida in the United States have been used in economic evaluations of folic acid fortification (Waitzman et al., 1994, 1996, 2005; Centers for Disease Control and Prevention, 1995; Romano et al., 1995; Kelly et al., 1996; Grosse et al., 2005). In addition to evaluating the effects of prevention strategies, such an understanding is necessary for developing cost-effective management strategies for both children and adults with spina bifida. Thus, we sought to estimate medical care expenditures among both children and working-age adults living with spina bifida.

## METHODS

### Data Source and Study Population

We obtained data from the MarketScan Commercial Claims and Encounters Database (CCE) for 2001–2003. The MarketScan CCE database consists of nationwide employer- and health plan-sourced data for several million individuals annually. There are >150 contributing employers, mostly U.S. Fortune 500 firms. Some 80 contributing health plans include a variety of fee-for-service (FFS), fully capitated, and partially capitated health plans. Although the MarketScan CCE database over-represents the South and females, it is one of the few data sources from which a large sample size can be attained for low-prevalence conditions. The database is constructed from paid medical and prescription drug claims of persons covered by employer-sponsored health insurance, including active employees, early retirees, and their dependents. This database captures person-specific clinical utilization, expenditures, and enrollment across inpatient, outpatient, and prescription drug services (MarketScan Database, 2003).

Inclusion criteria for the study population were as follows. First, we included persons who had complete coverage for 12 months during calendar year 2003. Inclusion of persons with partial-year coverage and annualized expenditures could have biased the estimate of annual expenditures if the partial-year medical care utilizations were not representative of medical care utilization over the course of a full year. Second, we included only those persons with enrollment in FFS plans because enrollees in capitated plans do not necessarily have reimbursements or accurate FFS equivalents recorded. Third, we included only those persons with prescription drug coverage in their insurance plan in order to ensure complete coverage of medical expenditures. Fourth, we included only those persons <65 years of age. Few persons ≥65 are included in the MarketScan CCE database as a result of primary coverage by Medicare.

### Identification of Spina Bifida

Enrollees with spina bifida were identified by International Classification of Disease, 9<sup>th</sup> Revision (ICD-9-CM) rubric "741" in their primary or secondary diagnosis from either inpatient admissions or outpatient visits claims. This ICD-9-CM rubric covers all forms of spina bifida aperta, including cases with or without hydrocephalus.

It excludes spina bifida occulta (ICD-9-CM 756.17). We went through the databases for 2001–2003, and any individual who was identified in any encounter during those 3 years as having spina bifida was classified as having spina bifida, regardless of whether an ICD-9-CM rubric 741 was recorded in 2003. Infants born in 2002 were classified as having spina bifida if they had an ICD-9-CM rubric 741 in either 2002 or 2003.

### Measures of Medical Care Utilization and Expenditures

We extracted data on inpatient admissions, outpatient visits, and prescription drug services of the study population from the 2003 MarketScan CCE database. Medical care utilization was captured by the number of inpatient admissions, the average length of stay per admission, the number of outpatient visits, and the number of prescription drug claims used in 2003. We assessed medical expenditures from the health care payer perspective by three categories: health insurance; patient out-of-pocket expenses, which included copayments, coinsurance, and deductibles; and other, which refers to the amount of any payments made to the provider from a source other than the patient and the plan that submitted the claim, such as the amount paid by a spouse's insurance. We also reported medical expenditures by site of care, including inpatient, outpatient, and prescription drugs.

### Age Groups

To fully capture changes in the medical care utilization and expenditure over the life span, we classified the study population in four age groups: infants (<1 year old), children (ages 1–17 years), young adults (ages 18–44 years), and older adults (ages 45–64 years). For the last three age groups, we used the age in years recorded in the enrollment file, which means that an individual was assigned to the same age group for an entire year. We used a more refined way to identify the 0-year age group so that we could capture their medical care utilization and expenditures for 12 months starting at birth. This is important because infants with spina bifida typically undergo major surgeries during the first few months of life. Use of the 0-year age group from the 2003 enrollment file who had a full year enrollment in 2003 was problematic because it consisted almost exclusively of children born during 2002, and their medical care during 2003 missed the first few months of life. Instead, we included infants whose birthdates could be identified from their mothers' delivery records during 2002 and for whom 12 months of subsequent data were available.

### Data Analysis

We calculated medical utilization and expenditures for persons with and without spina bifida, stratified by age, site of care, and source of payment. We compared total mean expenditures and site-specific mean expenditures and used the 2-tailed *t* test to determine whether differences were statistically significant. We followed established practice in interpreting the differences in medical expenditures between persons with spina bifida and persons without spina bifida as reflecting the incremental costs associated with spina bifida, including disease

Table 1  
Sample Distribution of Age, Sex, and Relationship to Main Beneficiary

	Full sample	Persons with spina bifida	Administrative prevalence per 10,000 enrollees
Total, <i>n</i>	4,076,854	1,609	3.95
By age in years			
0–17 (%)	22.7	32.6	5.65
18–44 (%)	36.0	40.5	4.44
45–64 (%)	41.3	26.9	3.57
Male (%)	46.6	39.5	
By relationship to main beneficiary			
Employee (%)	47.6	31.6	
Spouse (%)	23.9	18.4	
Dependent (%)	28.5	50.0	

Full study sample and the subset with spina bifida are given (MarketScan CCE Database, 2003).

sequelae and secondary conditions that result from spina bifida and associated congenital malformations (Waitzman et al., 1994, 1996, 2005; Centers for Disease Control and Prevention, 1995). The implicit assumption is that comorbidities among persons with spina bifida that are unrelated to spina bifida are not substantially more common than among persons without spina bifida.

We also investigated the effects of using single year versus multiple years of data to identify persons with spina bifida on the estimates of medical expenditures. Persons with spina bifida who do not receive medical care specific to spina bifida in a given year would be less likely to have an ICD-9-CM rubric for spina bifida recorded in connection with utilization of health care services. Consequently, restricting analysis to data on persons with a currently reported billing code of spina bifida could over-represent those with higher utilization of health care services.

All analyses were conducted using SAS software version 9.1 (SAS Institute, Cary, NC; SAS, 2003).

## RESULTS

The full sample consisted of 4,076,854 people <65 years of age enrolled in a FFS health insurance plan with prescription drug coverage for 365 days in 2003. Among them, there were 1,595 persons with spina bifida 1–64 years of age. They were selected from 2,312 persons identified with a spina bifida diagnosis in any year during 2001–2003 who were 1–64 years of age. Of the total of 2,312 persons with spina bifida recorded, 462 (20%) were excluded from the sample because they were in capitated plans. An additional 255 (11%) were excluded because of their partial-year coverage. Implications of these exclusions are discussed in the limitations section.

We also identified 14 infants born in 2002 who had a diagnosis of spina bifida in either 2002 or 2003 who enrolled in a FFS plan with prescription drug coverage for 12 months following birth. Together these 1,609 persons constituted our sample of persons with spina bifida for this study. The mean age of the 1,609 persons was 30 years.

Compared with the full sample, a larger proportion of persons with spina bifida were females and younger (Table 1). Persons with spina bifida were more likely to be included in the sample as dependents and spouses of the main beneficiary instead of being main beneficiaries

themselves. The administrative prevalence of spina bifida from our study is 3.95 per 10,000 enrollees, which is similar to the recently published national prevalence estimates, 3.68 per 10,000 live births (Canfield et al., 2005). Prevalence decreased with age in our study sample.

Table 2 compares medical care utilization of persons with spina bifida among infants, children, young adults, and older adults. On average, 24.4% of persons with spina bifida in FFS plans were hospitalized in a given year, with an average length of stay of 5.8 days. Infants with spina bifida differed from those in other age groups because all but one infant was rehospitalized in the 12 months following birth. Conditional on having at least one hospital admission not associated with delivery, infants with spina bifida averaged two hospital admissions in 2003. Most (97.1%) persons with spina bifida in our sample received outpatient care during 2003. This percentage did not vary appreciably by age. On average, 87.4% of the sample had a least one prescription drug claim in 2003. Prescription drug claims rose as persons with spina bifida aged, from an average of five claims for infants to an average of 37 claims for those 45–64 years of age in 2003.

Table 3 shows average gross medical expenditures by disposition (inpatient, outpatient, and prescription drug) and payee (insurance, patient out-of-pocket, and other) by age group. Infants incurred higher average total expenditures (\$49,602) than other age groups. Hospitalization accounted for much of the high expenditures during infancy, with average expenditures exceeding \$40,000. Excluding infants, average total medical expenditures for persons with spina bifida did not increase with age, exceeding \$15,000 for each age group in 2003. Outpatient expenditures were slightly higher for infants. In contrast, expenditures for prescription drugs increased with age. Insurance, patients, and other sources paid 83.5, 11, and 5.5% of total expenditures, respectively. Out of the average total expenditures of \$1,689 per person paid by families in 2003, 33.8% was for inpatient admissions, 49.4% was for outpatient visits, and 16.8% was for prescription drugs.

The incremental medical expenditures by persons with spina bifida are the differences in average medical care expenditures for persons with and those without spina bifida. The differences in average medical expenditures between those with and without spina bifida were significant in all categories across age groups (Table 4). For

Table 2  
Medical Care Utilization by Persons with Spina Bifida

	Age group (years)				
	0	1-17	18-44	45-64	All ages
Number of persons with spina bifida	14	510	652	433	1,609
Inpatient admissions					
Number of persons	13	110	173	97	393
Percent of persons with inpatient admissions	92.9%	21.6%	26.5%	22.4%	24.4%
Number of admissions	28	173	246	142	589
Mean number of days per hospital stay	5.9	5.5	6.0	5.9	5.8
Outpatient visits					
Number of persons	14	494	632	422	1,562
Percent of persons with outpatient care	100%	96.9%	96.9%	97.5%	97.1%
Number of visits	374	11,896	14,499	11,169	37,938
Average number of visits for sample	27	23	22	26	24
Prescription drug					
Number of persons	14	417	567	408	1,406
Percent of persons with drug claims	100%	81.8%	87.0%	94.2%	87.4%
Number of drug claims	60	6,363	13,319	15,952	35,694
Average number of drug claims for sample	5	12	20	37	22

MarketScan CCE Database (2003).

persons without spina bifida, medical expenditures on inpatient admissions, outpatient visits, and prescription drugs all increased with age after infancy. In comparison, for persons with spina bifida, the medical expenditures on inpatient admissions were highest during infancy and remained stable afterwards. Expenditures on outpatient visits among persons with spina bifida were stable throughout the life course, whereas expenditures on prescription drugs increased with age. The resulting incremental medical expenditures by persons with spina bifida decreased with age. We also reported the ratio of mean expenditures to characterize the relative impact of living with spina bifida. Both the absolute and relative impact of spina bifida on medical expenditures decreased with increasing age among adults.

To investigate how the results might differ depending on the case identification strategy, Table 5 provides a comparison of expenditures estimates generated using only 1 year of diagnosis versus 3 years of diagnosis, respectively. Of 1,595 persons with spina bifida 1-64 years of age in 2003, only 62.4% had a spina bifida diagnosis recorded in a medical appointment during 2003. Adults with spina bifida would have been under-represented if we had used diagnoses in 2003 only. Total medical expenditures would have been overestimated by ~30%. Both expenditures on inpatient admissions and outpatient visits would have been overestimated, by 45% and 26%, respectively. Expenditures on prescription drugs would have been underestimated by 13%. The comparison shows that for chronic conditions with differ-

Table 3  
Average Expenditures in 2003 Dollars by Sources of Payment for Persons with Spina Bifida

	Age group (years)				
	0	1-17	18-44	45-64	All ages
No. of persons	14	510	652	433	1,609
Inpatient admissions					
Insurance	\$37,045	\$5,943	\$6,375	\$3,667	\$5,776
Other	0	49	456	1,582	626
Patient out of pocket	3,168	470	758	326	571
Total	40,213	6,462	7,589	5,575	6,973
Outpatient visits					
Insurance	8,038	6,931	5,657	5,883	6,142
Other	0	122	284	573	308
Patient out of pocket	691	886	817	806	835
Total	8,729	7,939	6,758	7,262	7,285
Prescription drugs					
Insurance	560	678	1,264	2,372	1,370
Other	0	0	0	0	0
Patient out of pocket	100	163	258	469	283
Total	660	841	1,522	2,841	1,653
Total cost (standard deviation)	49,602 (53,150)	15,242 (32,590)	15,869 (36,597)	15,678 (33,254)	15,911 (34,536)

MarketScan CCE Database (2003).

Table 4  
Per Capita Medical Expenditures in 2003 Dollars: Persons with versus without Spina Bifida

Age groups (years)	Mean expenditure, persons with spina bifida	Mean expenditures, persons without spina bifida	Difference in means	95% CIs of the difference in means	P	Ratio of means
Inpatient admissions			\$34,736			
0	\$40,213	\$5,477		16,310-53,161	.0265	7.3
1-17	6,462	193	6,269	5,819-6,712	.0001	33.5
18-44	7,589	625	6,964	6,396-7,509	.0001	12.1
45-64	5,575	1,369	4,206	2,458-5,969	.0001	4.1
0	8,729	2,450	6,279	1,214-11,345	.0156	3.6
1-17	7,939	759	7,180	6,878-7,472	.0001	10.5
18-44	6,758	1,429	5,329	4,877-5,697	.0001	4.7
45-64	7,262	2,778	4,484	3,644-5,375	.0001	2.6
0	660	215	445	112-1,002	.3037	3.1
1-17	841	222	619	482-728	.0001	3.8
18-44	1,522	479	1,043	895-1,154	.0001	3.2
45-64	2,841	1,398	1,443	1,219-1,691	.0001	2.0
0	49,602	8,142	41,460	20,890-62,029	.0120	6.1
1-17	15,242	1,172	14,070	13,437-14,664	.0001	13.0
18-44	15,869	2,530	13,339	12,472-14,057	.0001	6.3
45-64	15,678	5,544	10,134	8,024-12,333	.0001	2.8
Prescription drugs						
0						
1-17						
18-44						
45-64						
Total						
0						
1-17						
18-44						
45-64						

MarketScan CCE Database (2003).

Table 5  
Effects on Medical Expenditures of Using Single Year versus Multiple Years of Diagnoses to Identify Persons with Spina Bifida

Age group (years)	Mean medical expenditures in 2003 dollars			
	No.	Inpatient	Outpatient	Prescription drugs Total
Diagnosis recorded in year 2003 only				
1-17	365	8,759	9,700	886 19,345
18-44	409	10,898	8,048	1,244 20,190
45-64	221	9,030	10,162	2,773 21,965
Diagnosis recorded in any year 2001-2003				
1-17	510	6,462	7,939	841 15,242
18-44	652	7,589	6,758	1,522 15,869
45-64	433	5,575	7,262	2,841 15,678

ent severities, expenditure estimates might be overestimated if a single year of diagnosis is used to identify persons with the condition. It also shows that persons with less severe chronic conditions tend to use more prescription drugs and less inpatient and outpatient services for their care.

### DISCUSSION

Spina bifida is a chronic condition that persons have to manage over a lifetime. Previous analyses reported that children and adolescents with spina bifida incur medical expenditures several times higher than those children or adolescents who do not have spina bifida (Waitzman et al., 1996; Ireys et al., 1997). Two other recent studies also indicate that young adults with spina bifida continue to be high users of medical care (Young et al., 2005; Okumura et al., 2006). Our study includes a sample of persons with spina bifida at different ages throughout the lifespan and compares their medical care utilization and expenditures at different stages of life. In our analysis, adults accounted for 67% of persons with spina bifida and 66% of medical expenditures associated with spina bifida during 2003. We also investigated the hospital discharge data from the Health Care Utilization Project, which indicates that 62% of hospital discharges with a diagnosis of spina bifida in the National Inpatient Sample in 2003 occurred among adults ≥18 years of age (Agency for Healthcare Research and Quality, 2006). Those findings appear consistent with our results.

Our findings also indicated that children with spina bifida incur high medical expenditures relative to other children. Compared with that of the general population, the highest expenditure ratio between persons with and those without spina bifida, 13.0, was found for children 1-17 years of age. In comparison, data from the 2000 Medical Expenditure Panel Survey showed that children with special health care needs on average incurred medical expenditure three times higher than other children (Newacheck et al., 2005). These results indicate that children with spina bifida incur higher than average medical care utilization and expenditures among children with special health care needs.

We also compared estimates from the present study with inflation-adjusted estimates based largely on 1988-1989 California data (Waitzman et al., 1996, 2005). The ratio of average medical expenditures between those

with spina bifida and those without spina bifida is almost identical in these two studies for children, 7.0 and 7.2, respectively, at 0–1 years and 13.0 and 12.5, respectively, at 2–17 years. In contrast, the ratio for adults with spina bifida 18–44 years of age relative to adults without spina bifida was 6.8 in the present study, compared with 1.6 for adults  $\geq 18$  years of age in the Waitzman study. Among persons with spina bifida, the ratio of average expenditures between adults and children 1–17 years of age was 0.72 to 0.95 in the present study, compared with 0.32 in the previous study.

The higher incremental medical expenditures for adults with spina bifida in the present study relative to the Waitzman study could reflect a number of factors. First, the medical expenditures for adults with spina bifida could be understated in the previous analysis due to their data limitations. Second, medical expenditures for adults without spina bifida could be understated in the MarketScan data if the employed population is healthier than the general working-age population. Third, the increase in recorded medical expenditures for adults with spina bifida could be real, reflecting improvements in treatment, especially surgery and physical therapy, as well as increased survival of individuals with severe forms of spina bifida who might have died previously. We are not able to choose among these different explanations on the basis of the present data, although we consider the most likely explanation to be incomplete coverage in the previous study of outpatient utilization and expenditures among adults with spina bifida for secondary conditions and comorbidities.

To explore the implications of the findings of the present study for estimates of lifetime expenditures, we calculated the present value of lifetime medical expenditures in the 2003 MarketScan data using the same life table survival probabilities used in the Waitzman study. The present value of lifetime incremental medical expenditures using a discount rate of 3% worked out to  $\sim$ \$319,000, which compares with an estimate of \$236,000 in 2003 dollars based on the 1988–1989 estimates adjusted for inflation. The difference in lifetime expenditure estimates reflect multiple factors, including differences between privately insured and publicly insured patients, changes in case mix and technology, and methods of cost adjustment. In order to determine the contribution of the difference in expenditure estimates for adults between these two studies, we calculated the present value using the ratio of average expenditures between adults and children from the Waitzman study, 0.32, in combination with our estimates for children. This yields a present value estimate of \$223,000, which is 30% lower than the \$319,000 estimate calculated using observed incremental expenditures for adults. The difference in adult expenditure estimates more than accounts for the difference in lifetime incremental medical expenditures between our estimates and the Waitzman estimates.

Despite these important findings, the study has a number of limitations. First, the sample is taken from an employment-based insured population, which is likely healthier and has better access to medical care than the general population. The analysis does not necessarily capture medical expenditures for those who had delayed and insufficient care. Thus, the results might not be generalizable to those with no coverage or less generous coverage.

Because of data limitations, we were unable to account for sample attrition due to persons switching to other types of insurance or employment. One important factor that could lead to differential sample attrition is severity. For example, children and adults with spina bifida who exhausted their lifetime private insurance allowance because of high utilization might have switched to public insurance such as Medicaid, making spina bifida less represented in this database in older age groups overall. An implication is that we have likely understated the share of adults with spina bifida. This fact only strengthens the conclusion that adults with spina bifida receive a large portion of all medical care for persons with spina bifida.

Another limitation is the restriction of the study to persons enrolled in FFS plans. Because of incomplete recording of FFS equivalent estimates of expenditures, we were unable to estimate medical costs for persons in capitated plans. Among the 20% of spina bifida patients enrolled in capitated plans the number of inpatient admissions was almost identical to those enrolled in FFS plans, although average length of stay was 25% less for the capitated plan sample (4.6 vs. 6.0 days;  $P < .001$ ). Although hospital expenditures were not necessarily proportional to length of stay, it is likely that inpatient expenditures were somewhat lower for spina bifida patients enrolled in capitated plans.

Finally, medical care expenditures constitute only a part of the total economic impact of spina bifida. Because the analysis was restricted to medical expenditures from administrative claims data, we did not capture the total cost of spina bifida. To do so would require information on medical care delivered outside health insurance plans, other out-of-pocket costs (such as medical durable equipment and the costs of residence modifications), costs of family caregiving, and human capital losses. How the magnitude of medical care expenditures compares with the total costs of spina bifida is a direction for future research.

In summary, for persons with spina bifida, average medical expenditures during the first year of life during 2002–2003 were  $\sim$ \$50,000. Inpatient admissions accounted for a large portion of total expenditures during infancy because surgeries are concentrated during infancy for persons with spina bifida. After infancy, average medical care expenditures per person with spina bifida during 2003 ranged from \$15,000 to \$16,000 per year among different age groups. Incremental expenditures associated with spina bifida were not stable, but decreased with increasing age, from  $\sim$ \$14,000 per year for children to  $\sim$ \$10,000 per year for adults 45–64 years of age.

We suggest that estimates of lifetime medical costs for spina bifida be updated based on the present findings on incremental medical care expenditures among adults with spina bifida. A new estimate of lifetime costs of spina bifida reflecting the changing demographics of the spina bifida population is necessary for developing cost-effective spina bifida management strategies for both children and adults. It would also allow more accurate evaluation of the effects of spina bifida prevention strategies.

In addition to quantifying medical expenditures for both children and adults with spina bifida, we used multiple years of diagnoses to identify persons with spina bifida. Previous studies that used claims diagnoses in

one given year to identify persons with a certain condition might have overestimated average costs if milder forms of the condition were not recorded on medical claims or if persons did not visit a medical provider in a given year. Whether this finding extends to other studies using MarketScan and other databases to quantify medical expenditures associated with disease is a direction for future work.

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