

# Health Care Utilization & Costs For Cystic Fibrosis Patients With Pulmonary Infections

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## ABSTRACT

**Purpose:** To examine patterns of health care utilization and costs among cystic fibrosis (CF) patients with pulmonary infections.

**Design:** Retrospective administrative claims database analysis.

**Methodology:** We used administrative claims data (including both medical and pharmacy claims) to examine health care utilization and costs among CF patients with pulmonary infections over one year. We conducted a subgroup analysis in which we examined selected outcome measures among patients with tobramycin for inhalation (TIS) prescriptions by the number of TIS prescriptions filled.

**Principal findings:** Among 1,064 CF patients identified with pulmonary infections, 80% had at least one CF-related office visit, 34% had a CF-related hospital stay, and 95% filled at least one prescription over one year. Total annual CF-related health care costs averaged \$29,000 plus \$20,000 for prescription drugs. In the subgroup analysis, there was a trend towards longer lengths of stay and higher inpatient costs with fewer numbers of TIS prescriptions filled.

**Conclusion:** CF patients with pulmonary infections have substantial

levels of health care utilization and costs.

## INTRODUCTION

Cystic fibrosis (CF) is an autosomal recessive disease caused by abnormalities of the cystic fibrosis transmembrane conductance regulator (CFTR) chloride conductance channel and affecting the function of exocrine glands from multiple organs, including the lungs, liver, pancreas, intestines, and skin. CF affects an estimated 100,000 people worldwide: approximately 50,000 in Europe and 30,000 in the United States (Boyle 2007, Cystic Fibrosis Worldwide. Annual report 2005, Cystic Fibrosis Foundation 2006, Cystic Fibrosis Worldwide. International 2007, Cystic Fibrosis Trust 2007, European Lung Foundation 2009, French National Cystic Fibrosis Observatory 2006). Despite the absence of curative therapy, advances in medical care have extended the median predicted age of survival for an individual with CF from less than 5 years in 1960 to nearly 40 years in 2006 (Cystic Fibrosis Foundation 2006).

Clinically, CF is characterized by malabsorption, gastrointestinal abnormalities, infertility, and severe progressive sinopulmonary disease. Although the disease affects multiple organ systems, pulmonary disease constitutes the greatest source of morbidity and mortality. Individuals with CF produce abnormally thick

mucus that obstructs the airways. The airways become a focus for chronic infections that compromise the day-to-day function of the individual, leading to airway destruction, progressive lung disease, and ultimately, to death from respiratory failure.

CF patients are particularly susceptible to pulmonary infections with *Pseudomonas aeruginosa*, (*PA*), a gram-negative, biofilm-producing pathogen that is ubiquitous in adult CF patients (Davis 1996). Chronic airway infection with *PA* is a significant predictor of mortality in CF patients (Henry 1992), and has also been associated with faster rates of pulmonary function decline (Pamukcu 1995). Once *PA* infection is established in the respiratory tract of a CF patient, the patient's clinical course can change dramatically (Cystic Fibrosis Trust Antibiotic Group 2002). A recent analysis showed an association between infection with *PA* and more severely compromised lung function (Pittman 2008), and loss of lung function is the primary cause of death in CF patients (Corey 1997). The Cystic Fibrosis Foundation Patient Registry, which includes data for about 80% of the known CF patients in the United States, attributed 69% of CF patient deaths to pulmonary function decline and related sequelae (Cystic Fibrosis Foundation 2006).

Recommended pharmacologic therapy under published guidelines for pulmonary complications for cystic fibrosis include dornase alfa (rhDNase), a mucolytic given daily; azithromycin, a macrolide antibiotic

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that has anti-inflammatory properties; and tobramycin inhalation solution (TIS), an inhaled antibiotic given in 28 day on/off cycles (Flume 2007). All three have been shown to improve lung function and to decrease the incidence of pulmonary exacerbations.

A chronic, costly disease like CF requires effective and efficient care management to ensure quality of care and help contain health care costs. Providers and payers will be increasingly at risk as reimbursement policies evolve in a changing health care environment, and efficient use of resources is essential for both patient outcomes and payers' expenditures. Treatment guidelines play a key role in evidence-based management of patients, and adherence to treatment guidelines is important for patient outcomes and quality of life. Although various treatments now exist to improve patient outcomes and quality of life in CF, and guidelines for use of these treatments have been established, the CF Foundation reports that only one-third of patients adequately comply with CF medications (Cystic Fibrosis Foundation 2006). If this is the case, there may be room for improved outcomes and potentially lower health care utilization and costs among CF patients. An analysis of the current health care utilization (including prescription drugs) and costs among CF patients in a commercially insured population in the U.S. can provide information to physicians and payers on the extent to which patients are complying with medications and treatment guidelines.

Although it is recognized that CF patients have high health care needs, particularly once pulmonary infections arise (Yankaskas 2004), very few studies have documented patterns of health care utilization and cost among these patients. Furthermore, existing studies have either focused on a specific subpopulation (Oster 1995) or are outdated (Oster 1995, Lieu 1999). As treatment patterns for CF have changed dramatically in the

past 10 years with the widespread adoption of all three above mentioned agents, an updated assessment of health care utilization and costs is warranted.

In this study, we used administrative claims data from a large, national U.S. health plan to examine health care utilization and costs over one year among CF patients with pulmonary infections. We stratified patients by age and compared outcomes between children and adults. We conducted a subgroup analysis among patients with at least one prescription for TIS, in which we examined selected outcomes by numbers of TIS prescriptions filled (i.e., 1–2, 3–5, or 6+) during the 12-month period.

## METHODS

### Overview

We used administrative claims data from a large U.S. health plan to evaluate health care utilization and costs among patients with CF with pulmonary infections. Patient characteristics (age, gender, health plan geography, length of follow-up) as well as measures of health care utilization and cost were analyzed separately for children (i.e., those aged 6–17 years of age) and adults (i.e., 18 years of age and older). Health care utilization and cost measures included office visits, outpatient hospital visits, emergency room (ER) visits, inpatient stays, and prescription drugs. Study measures were assessed over a 12-month follow-up period.

Using patients from the same cohort, a subgroup analysis was conducted among patients who had at least one prescription for TIS during the identification period. TIS is recommended therapy for CF patients over the age of 6 years with pulmonary *P. aeruginosa* infection (Flume 2007). The recommended dosing schedule for TIS is 28 days on therapy followed by 28 days off therapy; based on these recommendations, a fully compliant patient would

be expected to have filled 6 prescriptions in the 12-month period following the index prescription. We stratified patients by number of prescriptions filled (i.e., 1–2 prescriptions, 3–5 prescriptions, and 6+ prescriptions filled) in a 12-month period and examined selected measures by number of prescriptions category.

## Data source

We conducted our analysis using medical and pharmacy claims data for commercial and Medicaid enrollees from a large U.S. health plan affiliated with i3 Innovus, an Ingenix Inc. company. Membership in the health plan is geographically diverse across the United States (approximately 11% from Northeast region, 31% Midwest, 43% South, and 14% West), and beneficiaries have fully-insured coverage for physician, hospital, and prescription drug services. In calendar year 2006, the administrative claims database included data for approximately 14 million health plan enrollees with both medical and pharmacy benefits. Claims data are de-identified, and each enrollee is assigned a randomly generated unique identifier before being placed in the database. The study was conducted in accordance with established corporate guidelines for adherence to Health Insurance Portability and Accountability Act (HIPAA) privacy requirements. Review by an institutional review board was not sought.

## Study subjects

Patients were included in the study if they had at least one medical claim with a listed diagnosis of *cystic fibrosis with pulmonary manifestations* (International Classification of Diseases, Ninth Revision, Clinical Modification [ICD-9-CM] 277.02) between October 1, 2002 and April 30, 2006. The first date of the diagnosis during the identification period was defined as the patient's "index date." Subjects were required to be 6 years of

age or older as of the year of the index date and to have at least 12 months of continuous enrollment following the index date, unless there was evidence that disenrollment was due to death (i.e., discharge status on a hospital claim was equal to "dead"). Subjects who met the criteria were stratified by age for purposes of the analyses, where children were defined as those aged 6–17 years of age and adults were defined as those 18 years of age and older.

A subgroup of patients with at least one prescription for TIS during the identification period was identified. Patients were stratified by number of prescriptions filled during the 12-month follow-up period as follows: 1–2 prescriptions, 3–5 prescriptions, and 6+ prescriptions filled.

### Study measures

Study measures included patient characteristics (age, gender, health plan region), health care utilization, and health care costs. Measures of health care utilization and cost included physician office visits, outpatient hospital visits, ER visits, inpatient stays, and prescription drugs. Utilization and costs of physician office visits, outpatient hospital visits, ER visits, inpatient stays were examined for the treatment of cystic fibrosis (ICD-9-CM 277.xx) and for any reason. Utilization and costs of prescription drugs were examined for TIS; dornase alfa; azithromycin; antibiotics other than TIS or azithromycin; and any drug. As data were drawn from an administrative claims database from a large U.S. health care insurer, patient out-of-pocket costs were not included.

### Statistical analyses

Patient characteristics were examined as of the index date. Health care utilization and cost measures were analyzed over the 12-month follow-up period. All study variables were analyzed descriptively and patient characteristics and health care uti-

lization and cost measures were compared across age groups using a t-test or chi-square test, as appropriate. For measures of health care utilization, proportions of patients utilizing services and the overall mean numbers of annual visits, stays, and prescriptions were calculated, along with standard deviations. Mean health care costs and standard deviations are presented on an annual basis across all patients.

In the subgroup analysis, patient characteristics were summarized by category of TIS prescriptions filled (i.e., 1–2 prescriptions, 3–5 prescriptions, and 6+ prescriptions). Characteristics were compared across categories using a t-test or chi-square test, as appropriate. Analyses of health care utilization focused on mean inpatient length of stay and mean cost per inpatient stay. These outcomes were compared across number of prescriptions category using both a t-test and a nonparametric Wilcoxon Rank Sum test.

## RESULTS

### Demographic characteristics

Within the study timeframe of October 1, 2002 and April 30, 2006, 1,064 patients met the selection criteria. Demographic characteristics and mean length of follow-up for children and adult patients with CF with pulmonary manifestations are displayed in Table 1. About 51% of patients in both groups were male; geographic distribution of the health plans was similar across groups. Average ages were 11.4 years in the pediatric group and 31.8 years in the adult group. Mean length of follow-up was slightly longer for the pediatric (363.2 days) versus the adult (357.4 days) population ( $p=0.0074$ ).

### Health care utilization

Utilization of health care services for the treatment of CF and any diagnosis in this population is reported in Figure 1 and Table 2. Relative to adults, higher proportions of chil-

dren had at least one CF-related office visit ( $p=0.0046$ ), CF-related outpatient hospital visit ( $p<0.0001$ ), outpatient hospital visit for any reason ( $p=0.0016$ ), and CF-related ER visit ( $p=0.0159$ ) (Figure 1). Percentages of patients with a prescription filled for TIS ( $p=0.0023$ ) and dornase alfa ( $p<0.001$ ) were also slightly higher among children than adults (data not shown).

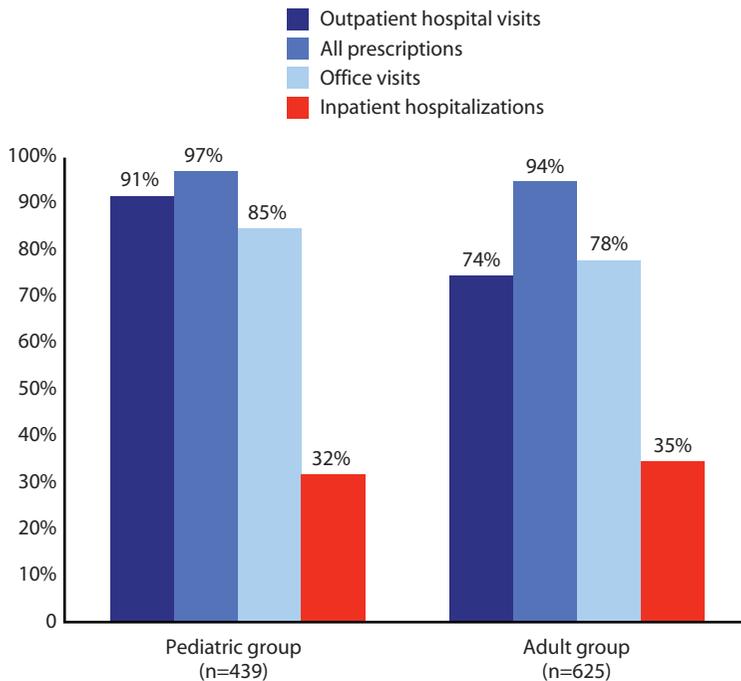
With respect to frequency of use, adults with CF averaged about 12 office visits per year for any diagnosis, compared to about 10 visits per year among children ( $p=0.0067$ ) (Table 2). About one-third of adult visits were specifically related to CF, compared to less than one-half of all visits among children. Children had more CF-related outpatient hospital visits ( $p=0.004$ ) as well as prescriptions for TIS ( $p=0.0007$ ) and dornase alfa ( $p<0.0001$ ) relative to adult patients. Adults had more frequent inpatient stays for any diagnosis ( $p=0.0021$ ) and numbers of prescriptions for antibiotics other than TIS and azithromycin relative to their younger counterparts ( $p=0.0009$ ). Adults also had an average of 43 prescriptions per year, which is more than pediatric patients (with 39 prescriptions) ( $p=0.03$ ).

### Health care costs

Health care costs (as billed by providers to the health plan) over the 12-month follow-up period are reported in Table 3. In general, mean costs of services did not differ significantly between children and adults; key exceptions to this were mean costs for dornase alfa prescriptions (higher among children,  $p<0.0001$ ) and azithromycin prescriptions (higher among adults,  $p<0.0001$ ). Total drug costs were higher among children than adult CF patients ( $p=0.0096$ ). In general, health care costs were high in this population, with mean annual costs for CF totaling about \$29,000 per patient plus another \$20,000 for prescription drugs. Mean annual

**TABLE 1****Patient characteristics, by age group**

Characteristic	Total (N=1,064)		Aged 6–17 years (N=439)		Aged 18+ years (N=625)		P-value
<b>Age group, years [N (%)]</b>							
< 18	<b>439</b>	(41.3)	<b>439</b>	(100.0)	<b>N/A</b>	N/A	N/A
18–34	<b>420</b>	(39.5)	<b>N/A</b>	N/A	<b>420</b>	(67.2)	N/A
35–64	<b>196</b>	(18.4)	<b>N/A</b>	N/A	<b>196</b>	(31.4)	N/A
65+	<b>9</b>	(0.9)	<b>N/A</b>	N/A	<b>9</b>	(1.4)	N/A
<b>Age [Mean (SD)]</b>	<b>23.39</b>	(13.7)	<b>11.35</b>	(3.4)	<b>31.84</b>	(11.8)	<.0001
<b>Gender [N (%)]</b>							
Male	<b>539</b>	(50.7)	<b>223</b>	(50.8)	<b>316</b>	(50.6)	0.9393
Female	<b>525</b>	(49.3)	<b>216</b>	(49.2)	<b>309</b>	(49.4)	0.9393
<b>Health plan region [N (%)]</b>							
Northeast	<b>118</b>	(11.1)	<b>48</b>	(10.9)	<b>70</b>	(11.2)	0.8918
Midwest	<b>378</b>	(35.5)	<b>143</b>	(32.6)	<b>235</b>	(37.6)	0.0917
South	<b>402</b>	(37.8)	<b>181</b>	(41.2)	<b>221</b>	(35.4)	0.0519
West	<b>166</b>	(15.6)	<b>67</b>	(15.3)	<b>99</b>	(15.8)	0.7981
<b>Length of follow-up, days [mean (SD)]</b>	<b>359.8</b>	(38.8)	<b>363.2</b>	(22.2)	<b>357.4</b>	(46.9)	0.0074

**FIGURE 1****Percent of patients by utilization**

health care costs for any diagnosis were over \$40,000. Inpatient hospital costs were particularly high, averaging \$22,102 for CF-related stays and \$26,761 for stays for any diagnosis. CF-related hospital costs comprised 75% of the total costs for CF-related services (excluding medications).

**Subgroup analysis**

A total of 577 (57%) patients had at least one prescription filled for TIS during the identification period. In the 12-month period following the first prescription for TIS, 278 (48%) patients had 1–2 prescriptions filled, 192 (33%) had 3–5 filled, and 107 (19%) had 6 or more prescriptions filled for TIS. Characteristics of these patients are contained in Table 4. The mean ages, gender distributions, and distributions of the health plans across the U.S. were comparable across the three TIS groups.

Patients with 6 or more prescriptions had complete follow-up (i.e., 365 days); mean lengths of follow-

**TABLE 2**  
Average health-care utilization per patient during the follow-up period

Utilization measure	Total (N=1,064) [Mean (SD)]		Aged 6–17 years (N=439) [Mean (SD)]		Aged 18+ years (N=625) [Mean (SD)]		P-value
Office visits							
CF-related	<b>4.39</b>	(7.77)	<b>4.76</b>	(10.14)	<b>4.12</b>	(5.52)	0.2283
Any diagnosis	<b>11.28</b>	(12.58)	<b>10.03</b>	(12.19)	<b>12.16</b>	(12.79)	0.0067
Outpatient hospital visits							
CF-related	<b>6.26</b>	(7.57)	<b>7.21</b>	(6.86)	<b>5.59</b>	(7.96)	0.0004
Any diagnosis	<b>11.75</b>	(13.18)	<b>11.05</b>	(9.99)	<b>12.24</b>	(15.01)	0.1209
ER visits							
CF-related	<b>0.19</b>	(1.81)	<b>0.33</b>	(2.76)	<b>0.09</b>	(0.47)	0.0727
Any diagnosis	<b>0.56</b>	(2.56)	<b>0.55</b>	(3.01)	<b>0.57</b>	(2.19)	0.9056
Inpatient hospitalization							
CF-related	<b>0.64</b>	(1.26)	<b>0.6</b>	(1.26)	<b>0.68</b>	(1.26)	0.2995
Any diagnosis	<b>0.84</b>	(1.43)	<b>0.69</b>	(1.35)	<b>0.95</b>	(1.47)	0.0021
Prescription drugs							
Tobramycin inhalation solution	<b>1.80</b>	(2.25)	<b>2.08</b>	(2.35)	<b>1.6</b>	(2.16)	0.0007
Dornase alfa	<b>3.09</b>	(4.09)	<b>3.96</b>	(4.19)	<b>2.47</b>	(3.91)	<.0001
Azithromycin	<b>2.65</b>	(4.52)	<b>2.56</b>	(4.75)	<b>2.71</b>	(4.35)	0.5997
Other antibiotics*	<b>7.46</b>	(9.73)	<b>6.36</b>	(7.54)	<b>8.24</b>	(10.95)	0.0009
Any drug	<b>41.25</b>	(34.01)	<b>38.71</b>	(29.89)	<b>43.04</b>	(36.55)	0.0344

\*Excluding tobramycin inhalation solution, dornase alfa, and azithromycin

up for the other two groups were only slightly shorter (360 days for 1–2 prescriptions and 364.4 for 3–5 prescriptions).

Number of CF-related hospitalizations, length of stay, and inpatient costs per stay are also contained in Table 4. While not statistically significantly different across the three groups, there was a trend towards more inpatient stays with longer lengths of stay and higher mean inpatient costs per stay as number of prescriptions filled for TIS decreased (Table 4).

## DISCUSSION

In this study, we used administrative claims data from a large U.S. health plan to quantify health care utilization and costs among patients with CF with pulmonary infections.

Health care utilization and costs were tabulated separately for children and adults over a 12-month follow-up period and included office visits, outpatient hospital visits, ER visits, inpatient stays, and prescription drugs. In addition, we conducted an alternative analysis among patients who had at least one prescription for TIS during the identification period. We stratified patients by number of prescriptions filled (i.e., 1–2, 3–5, and 6+ prescriptions filled in a 12-month period) and examined selected measures by number of prescriptions category.

Results of our study suggest that CF patients with pulmonary infections have high rates of health care utilization and costs. In general, outcomes did not vary significantly between children and adults, with a few

exceptions. The average number of office visits per year among all patients was 11 visits and the average number of outpatient hospital visits was 12 visits. CF patients have monthly contact with physicians. Despite this contact and management by physicians, 33% had at least one inpatient stay. There were, on average, 0.56 ER visits, 0.84 inpatient stays and 41 prescriptions per year for any diagnosis. Total annual health care costs were \$29,000 for CF-related (non-drug) services and \$40,000 for any diagnosis. Hospital costs comprised 75% of the total cost for CF-related services. The mean cost of medications among CF patients totaled an additional \$20,000 per year.

Results of the subgroup analysis indicate that a substantial proportion of patients receiving TIS are not re-

ceiving the drug as recommended. Because pulmonary infections are chronic among this population, administration of TIS is recommended in 28-day on and 28-day off treatment cycles, meaning that over a one-year period, patients should be filling 6 prescriptions for the drug. Our results indicate that only 19% of patients identified with an index prescription for TIS filled 6 or more prescriptions over the following one-year period. Over 48% of patients only filled 1 or 2 prescriptions, and over 33% filled 3–5 prescriptions. While the differences were not significant, we also found that numbers of in-

patient stays, lengths of stay, and inpatient costs all trended higher among patients with fewer prescriptions for TIS. This may indicate that reduced compliance with treatment guidelines results in worse outcomes and higher health care utilization and costs.

According to the CF Foundation, only one-third of patients adequately comply with CF medications. Although the CF guidelines strongly recommend chronic use of TIS in moderate and severe patients, only 62% of moderate-to-severe CF patients on TIS comply with clinical care guidelines (Cystic Fibrosis Foun-

ation 2006). Despite advances made in CF treatments, compliance to treatment is low and patients on current standard of care experience a 2% decline in pulmonary function annually (Konstan 2007). It is possible that more simplified drug delivery systems and treatment regimens may improve compliance among CF patients and thereby also patient clinical and economic burden.

Our study is one of the first to quantify the high health care needs among this population in the United States. Rosenberg & Farrell examined the costs of a CF diagnosis and subsequent treatment among children

**TABLE 3**  
**Average health-care cost per patient during the follow-up period**

<b>Cost measure</b>	<b>Total (N=1,064) [Mean (SD)]</b>		<b>Aged 6–17 Years (N=439) [Mean (SD)]</b>		<b>Aged 18+ Years (N=625) [Mean (SD)]</b>		<b>P-value</b>
Office visits							
CF-related	<b>625</b>	(1,255)	<b>585</b>	(615)	<b>652</b>	(1,555)	0.3337
Any diagnosis	<b>1,578</b>	(2,955)	<b>1,324</b>	(3,205)	<b>1,757</b>	(2,755)	0.0219
Outpatient hospital visits							
CF-related	<b>2,153</b>	(3,644)	<b>2,455</b>	(3,317)	<b>1,941</b>	(3,845)	0.0201
Any diagnosis	<b>5,401</b>	(11,229)	<b>4,767</b>	(8,548)	<b>5,846</b>	(12,767)	0.0992
ER visits							
CF-related	<b>22</b>	(144)	<b>34</b>	(200)	<b>13</b>	(85)	0.0457
Any diagnosis	<b>135</b>	(566)	<b>120</b>	(467)	<b>145</b>	(626)	0.4675
Inpatient hospitalization							
CF-related	<b>22,102</b>	(81,642)	<b>20,502</b>	(74,680)	<b>23,225</b>	(86,238)	0.5831
Any diagnosis	<b>26,761</b>	(87,288)	<b>22,247</b>	(76,499)	<b>29,931</b>	(94,060)	0.1431
Prescription drugs							
Tobramycin inhalation solution	<b>5,760</b>	(7,444)	<b>6,538</b>	(7,655)	<b>5,213</b>	(7,248)	<0.0042
Dornase alfa	<b>5,771</b>	(7,412)	<b>7,238</b>	(7,379)	<b>4,741</b>	(7,266)	<.0001
Azithromycin	<b>451</b>	(799)	<b>329</b>	(668)	<b>536</b>	(869)	<.0001
Other antibiotics*	<b>2,453</b>	(5,674)	<b>1,648</b>	(4,492)	<b>3,020</b>	(6,316)	<.0001
Any drug	<b>20,054</b>	(17,848)	<b>21,710</b>	(16,606)	<b>18,891</b>	(18,596)	0.0096
Total medical costs†							
CF-related	<b>29,143</b>	(84,943)	<b>27,597</b>	(77,990)	<b>30,228</b>	(89,550)	0.6106
Any diagnosis	<b>40,132</b>	(97,531)	<b>33,728</b>	(84,090)	<b>44,630</b>	(105,792)	0.0619

\*Excluding tobramycin inhalation solution and azithromycin.

†Excludes prescription drugs

**TABLE 4****Characteristics and outcomes among CF patients with a prescription for TIS during the identification period, by number of TIS prescriptions over 12 months**

Characteristic	1-2 Rx (N=278)	3-5 Rx (N=192)	6+ Rx (N=107)	P-value
Age group, years [N (%)]				
< 18	118 (42.5)	90 (46.9)	55 (51.4)	0.2602
18-34	119 (42.2)	76 (39.6)	39 (36.5)	0.4947
35-64	41 (14.8)	26 (13.5)	13 (12.2)	0.7937
65+	0 (0.0)	0 (0.0)	0 (0.0)	N/A
Age [mean (SD)]	21.7 (11.3)	21.1 (12.1)	20 (10.9)	0.4514
Gender [N (%)]				
Male	147 (52.9)	98 (51.0)	58 (54.2)	0.8588
Female	131 (47.1)	94 (49.0)	49 (45.8)	0.8588
Health plan region [N (%)]				
Northeast	25 (9.0)	21 (10.9)	11 (10.3)	0.7764
Midwest	93 (33.5)	70 (36.5)	43 (40.2)	0.4498
South	119 (42.8)	82 (42.7)	37 (34.6)	0.2996
West	41 (14.8)	19 (9.9)	16 (15.0)	0.2589
Length of follow-up, days [mean (SD)]	360.0 (39.0)	364.4 (8.7)	365.0 (0.0)	0.1282
Number of CF-related inpatient stays [mean (SD)]	1.9 (1.7)	1.8 (1.4)	1.5 (0.7)	0.42
Length of stay [mean (SD)]	8.8 (9.8)	8.2 (5.6)	7.2 (4.8)	0.55
Inpatient costs [mean (SD)]	\$67,864 (147,892)	\$52,422 (83,312)	\$36,636 (46,258)	0.36

with CF, but this study was comprised of data from the state of Wisconsin only (Rosenberg 2005). Oster (1995) examined the impact of recombinant human DNase on the cost of treating respiratory infections among CF patients who participated in a clinical trial (Oster 1995). Authors of this study reported that patients treated with DNase had lower rates of health care utilization and costs than those who were not treated with DNase. This study pertains to a selected group of patients from a trial; in addition, the data are now more than 10 years old. Lieu (1999) evaluated costs of CF patients in a health maintenance organization and reported health care costs for children with CF ranging from \$6,200 to \$43,300 per year for those with mild-to-severe

disease (Lieu 1999). However, this analysis was also done with data from over 10 years ago, and treatment patterns for CF have changed since then. Most notably, since the approval of TIS for pulmonary infections in 1998, outcomes among these patients have improved substantially. Our analysis therefore represents a more recent picture of health care utilization and costs among CF patients with pulmonary infections, conducted within a geographically diverse population in the United States

#### Limitations

Our study is not without limitations, primarily related to the use of administrative claims data. First, we identified patients based on ICD-9-CM codes contained in diagnosis

fields; patient identification was therefore subject to coding inaccuracies. Further, health care needs are likely to increase with increasing severity among this population. However, we were not able to capture lung disease severity among the CF patients in the claims database; we therefore stratified by age and compared outcome measures between children and adults. Age may be considered a loose proxy for severity as the majority of children are in the normal and mild lung disease severity categories; whereas the majority of adults have moderate or severe lung disease (Cystic Fibrosis Worldwide 2005).

Although the claims database represents a geographically diverse population in the United States, the ex-

tent to which our results can be generalized across the United States is unknown. Specifically, while all regions of the U.S. are represented in our study, most of our study population is located in the Midwest (35.5%) and South (37.8%). It is unknown to what extent variations in practice patterns and costs limit the generalizability of our findings.

## CONCLUSION

This analysis represents one of the few existing studies of health care utilization and costs among CF patients with pulmonary infections. Results of our study suggest that rates of health care utilization and costs among this population are substantial, with CF-related costs totaling over \$29,000 per year, and services for any diagnosis exceeding \$40,000 annually. Prescription drug costs add an additional \$20,000 per year to total costs among these patients. Additional research on the health and economic burden of CF on patients and families is warranted; in the meantime, there are opportunities for improved treatment algorithms for patients with this condition.

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