Out-of-pocket Medical Costs for Parents with Children with Down Syndrome in the United States

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BACKGROUND

- As prenatal testing for Down syndrome (DS) evolves, expectant parents must make decisions about their pregnancy options with limited time and information[1]
- Cuckle et al. estimated the incremental lifetime cost of raising a child with DS to be as high as $900,000 (2013 USD)[2]; this study used estimates from Waitzman et al.[3] which included direct costs and indirect costs over the entire lifespan for 558 individuals born in California in 1998 with DS
- Boulet et al. have also estimated that the annual average incremental medical costs incurred by insurers for children with DS under 5 years of age was approximately $33,347 (2004 USD)[4]; their study does not address individual out-of-pocket costs to parents
- Expectant couples making pregnancy decisions are more interested in knowing what their individual out-of-pocket expenses for raising a child with DS will be rather than societal costs or medical costs incurred by their insurance company
- Little information about costs from the individual parental perspective exists in the current literature

STUDY OBJECTIVE

To estimate the out-of-pocket health care costs associated with raising a child with DS between birth and 18 years of age from the perspective of commercially insured parents

METHODS

Data

- Data from the OptumHealth Reporting and Insights employer-based claims database were used to conduct this retrospective cohort study
  - The database contains administrative claims (medical claims and pharmacy claims) and eligibility information for over 18 million individuals who are privately insured through their employers, including primary subscribers and their covered beneficiaries
  - Database covers the period from the first quarter of 1999 through the first quarter of 2013 and has been cited in many peer-reviewed publications on medical costs[5,6,7]

Selection criteria

- Patients were selected to be included in the study if they were enrolled in their family insurance plan as a child (<18 years of age); had an identifiable mother and/or father on the insurance plan; and had discernable demographic, enrollment, and parental characteristics to use in the matching algorithm (**Figure 1**)
  - DS case group: patients with at least one medical claim associated with a diagnosis of DS (ICD-9-CM code: 758.0x)
  - Control group: patients without any diagnoses for chromosomal conditions (ICD-9-CM code: 758.xx)

Observation time

- Each patient's observation time, during which he or she was enrolled in an insurance plan, was divided into clinically relevant age categories based on the American Academy of Pediatrics health care guides for patients with DS (**Figure 2**)[8]
  - <1 year old
  - 1 to <3 years old
  - 3 to <5 years old
  - 5 to <13 years old
  - 13 to <18 years old
- Patients were assigned to one or more age categories based on their enrollment period
- The first continuous enrollment period was used in the analysis for patients with multiple continuous enrollment periods

Figure 1. Sample selection

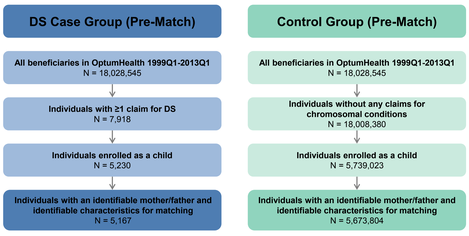
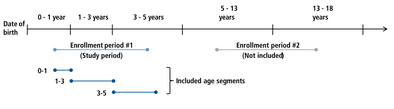


Figure 2. Study scheme



Patient matching

- Patients within each age category in the DS case cohort were matched to control patients in the same age category in a 1:4 ratio using a greedy matching algorithm[9,10]
  - Exact matching was used to match patient-age observations on gender, length of continuous eligibility, calendar year, age, and average age of parents at birth, while a propensity score was calculated to match patients on other covariates

Baseline characteristics, outcomes, and statistical analysis

- Baseline characteristics
  - Patient characteristics: age, sex, region, insurance plan type
  - Family characteristics: identifiable mother or father, average age of parents at patient's birth, parent work industry
  - Years of continuous follow-up
- Study outcomes
  - Average out-of-pocket annual health care utilization costs (i.e. co-pay and co-insurance payments) for each age category
- Analyses
  - Average yearly health care utilization costs were compared between the two cohorts using Wilcoxon signed-rank tests

RESULTS

Baseline characteristics post-matching

- After matching, patient age-cohorts were statistically similar with respect to most baseline characteristics (**Table 1**)

Table 1. Baseline characteristics in patient age cohorts, after matching

	Age 0 to <1			Age 1 to <3			Age 3 to <5			Age 5 to <13			Age 13 to <18		
	DS case group (N = 1,371)	Control group (N = 5,484)	*P*-Value[1]	DS case group (N = 1,457)	Control group (N = 5,828)	*P*-Value[1]	DS case group (N = 1,407)	Control group (N = 5,628)	*P*-Value[1]	DS case group (N = 2,126)	Control group (N = 8,504)	*P*-Value[1]	DS case group (N = 1,334)	Control group (N = 5,336)	*P*-Value[1]
Descriptive characteristics															
Male, n (%)	721 (52.6%)	2,884 (52.6%)	1.000	778 (53.4%)	3,112 (53.4%)	1.000	774 (55.0%)	3,096 (55.0%)	1.000	1,179 (55.5%)	4,716 (55.5%)	1.000	711 (53.3%)	2,844 (53.3%)	1.000
Age at earliest enrollment (years), mean ± SD	0.11 ± 0.23	0.11 ± 0.23	0.033 *	0.72 ± 0.92	0.72 ± 0.91	0.240	1.81 ± 1.65	1.81 ± 1.65	0.983	6.03 ± 3.93	6.05 ± 3.92	0.005 *	11.97 ± 4.10	11.98 ± 4.10	0.544
Region, n (%)															
Northeast	225 (16.4%)	921 (16.8%)	0.525	255 (17.5%)	1,192 (20.5%)	<0.001 *	246 (17.5%)	1,156 (20.5%)	<0.001 *	353 (16.6%)	1,674 (19.7%)	<0.001 *	245 (18.4%)	960 (18.0%)	0.539
Midwest	334 (24.4%)	1,284 (23.4%)	0.198	363 (24.9%)	1,329 (22.8%)	<0.001 *	345 (24.5%)	1,249 (22.2%)	<0.001 *	523 (24.6%)	1,889 (22.2%)	<0.001 *	308 (23.1%)	1,181 (22.1%)	0.170
South	499 (36.4%)	1,963 (35.8%)	0.471	517 (35.5%)	1,937 (33.2%)	0.004 *	504 (35.8%)	1,967 (35.0%)	0.273	721 (33.9%)	2,756 (32.4%)	0.017 *	405 (30.4%)	1,583 (29.7%)	0.146
West	232 (16.9%)	974 (17.8%)	0.138	223 (15.3%)	963 (16.5%)	0.020 *	211 (15.0%)	860 (15.3%)	0.587	362 (17.0%)	1,498 (17.6%)	0.263	238 (17.8%)	1,059 (19.8%)	0.002 *
Unknown	81 (5.9%)	342 (6.2%)	0.109	99 (6.8%)	407 (7.0%)	0.347	101 (7.2%)	396 (7.0%)	0.524	167 (7.9%)	687 (8.1%)	0.129	138 (10.3%)	553 (10.4%)	0.919
Insurance Plan Type, n (%)															
HMO	174 (12.7%)	691 (12.6%)	0.840	177 (12.1%)	669 (11.5%)	0.097	172 (12.2%)	761 (13.5%)	0.001 *	235 (11.1%)	981 (11.5%)	0.095	161 (12.1%)	678 (12.7%)	0.130
Indemnity	90 (6.6%)	384 (7.0%)	0.120	101 (6.9%)	396 (6.8%)	0.631	95 (6.8%)	380 (6.8%)	1.000	149 (7.0%)	598 (7.0%)	0.932	116 (8.7%)	496 (9.3%)	0.144
POS	255 (18.6%)	1,142 (20.8%)	0.001 *	266 (18.3%)	1,089 (18.7%)	0.501	265 (18.8%)	1,096 (19.5%)	0.315	433 (20.4%)	1,761 (20.7%)	0.473	296 (22.2%)	1,232 (23.1%)	0.185
PPO	777 (56.7%)	2,946 (53.7%)	<0.001 *	835 (57.3%)	3,376 (57.9%)	0.394	801 (56.9%)	3,073 (54.6%)	0.001 *	1,205 (56.7%)	4,811 (56.6%)	0.846	696 (52.2%)	2,674 (50.1%)	0.007 *
Other	75 (5.5%)	321 (5.9%)	0.341	78 (5.4%)	298 (5.1%)	0.505	74 (5.3%)	318 (5.7%)	0.329	104 (4.9%)	353 (4.2%)	0.004 *	65 (4.9%)	256 (4.8%)	0.840
Family characteristics															
Average age of parents at individual's birth (years), mean ± SD	35.59 ± 5.66	35.58 ± 5.64	0.003 *	35.33 ± 5.61	35.32 ± 5.59	0.005 *	35.10 ± 5.59	35.09 ± 5.57	0.101	33.87 ± 5.81	33.85 ± 5.78	<0.001 *	32.10 ± 5.73	32.09 ± 5.71	0.006 *
Parent work industry, n (%)															
Financial Services	156 (11.4%)	601 (11.0%)	0.455	187 (12.8%)	750 (12.9%)	0.950	178 (12.7%)	724 (12.9%)	0.691	240 (11.3%)	1,049 (12.3%)	0.008 *	118 (8.8%)	477 (8.9%)	0.812
Healthcare	90 (6.6%)	371 (6.8%)	0.654	114 (7.8%)	398 (6.8%)	0.025 *	90 (6.4%)	306 (5.4%)	0.016 *	121 (5.7%)	522 (6.1%)	0.128	83 (6.2%)	338 (6.3%)	0.764
Manufacturing/Energy	172 (12.5%)	682 (12.4%)	0.844	188 (12.9%)	808 (13.9%)	0.097	194 (13.8%)	726 (12.9%)	0.118	311 (14.6%)	1,197 (14.1%)	0.225	214 (16.0%)	844 (15.8%)	0.701
Retail/Consumer Goods	137 (10.0%)	475 (8.7%)	0.008 *	137 (9.4%)	474 (8.1%)	0.004 *	116 (8.2%)	365 (6.5%)	<0.001 *	153 (7.2%)	547 (6.4%)	0.004 *	87 (6.5%)	321 (6.0%)	0.115
Shipping/Transportation	305 (22.2%)	1,233 (22.5%)	0.719	305 (20.9%)	1,212 (20.8%)	0.833	306 (21.7%)	1,240 (22.0%)	0.654	452 (21.3%)	1,847 (21.7%)	0.343	270 (20.2%)	1,155 (21.6%)	0.020 *
Technology	362 (26.4%)	1,507 (27.5%)	0.128	358 (24.6%)	1,488 (25.5%)	0.136	360 (25.6%)	1,528 (27.1%)	0.022 *	581 (27.3%)	2,233 (26.3%)	0.035 *	354 (26.5%)	1,361 (25.5%)	0.071
Other	149 (10.9%)	615 (11.2%)	0.361	168 (11.5%)	698 (12.0%)	0.227	163 (11.6%)	739 (13.1%)	<0.001 *	268 (12.6%)	1,109 (13.0%)	0.077	208 (15.6%)	840 (15.7%)	0.566

Notes: **P*-value <0.05. [1] *P*-values were calculated using Wilcoxon signed-rank tests for continuous variables and McNemar tests for categorical variables.

Out-of-pocket costs

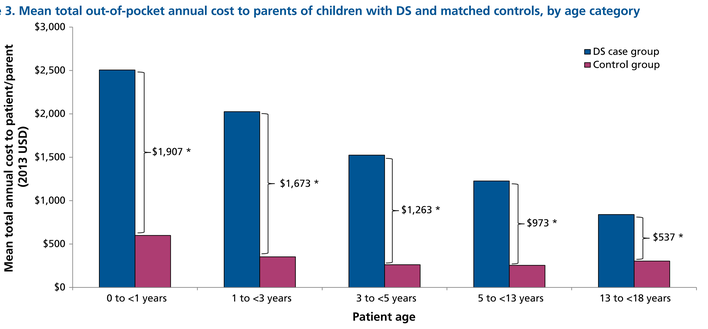
- Parents of patients with DS had significantly higher average annual out-of-pocket costs compared to their matched controls in each age group and cost category (**Table 2**)
- The greatest incremental out-of-pocket costs were inpatient costs during the first year of life ($1,183 vs. $259, $p<0.001$) and outpatient costs in later years of life (incremental costs range from $183 to $623, $p<0.001$)
- Mean total annual incremental out-of-pocket costs are highest for patients with DS from birth to 1 year of age ($1,907, $p<0.001$) (**Figure 3**)
- Patients with DS incurred incremental out-of-pocket medical costs of $18,248 from birth to 18 years of age

Table 2. Mean annual cost (to patient/parent) among individuals with DS and matched controls, by age category[1]

	DS case group	Control group	Cost difference	*P*-Value[2]
Age 0 to <1				
Patients	N = 1,371	N = 5,484	-	-
Total annual cost, mean ± SD	$2,506 ± $3,721	$599 ± $1,579	$1,907	<0.001 *
Total medical cost	$2,357 ± $3,647	$551 ± $1,549	$1,806	<0.001 *
Inpatient	$1,183 ± $2,888	$259 ± $1,346	$925	<0.001 *
Outpatient	$705 ± $946	$232 ± $439	$473	<0.001 *
ER	$106 ± $710	$44 ± $211	$63	<0.001 *
Home health agency	$335 ± $1,133	$14 ± $175	$322	<0.001 *
Other	$27 ± $173	$4 ± $33	$23	<0.001 *
Total pharmacy cost	$149 ± $384	$47 ± $131	$102	<0.001 *
Age 1 to <3				
Patients	N = 1,457	N = 5,828	-	-
Total annual cost, mean ± SD	$2,026 ± $3,354	$353 ± $801	$1,673	<0.001 *
Total medical cost	$1,826 ± $3,123	$305 ± $768	$1,521	<0.001 *
Inpatient	$480 ± $1,334	$64 ± $503	$416	<0.001 *
Outpatient	$796 ± $1,228	$173 ± $342	$623	<0.001 *
ER	$76 ± $271	$52 ± $242	$23	<0.001 *
Home health agency	$442 ± $1,894	$11 ± $103	$431	<0.001 *
Other	$33 ± $203	$5 ± $111	$27	<0.001 *
Total pharmacy cost	$201 ± $934	$48 ± $107	$153	<0.001 *
Age 3 to <5				
Patients	N = 1,407	N = 5,628	-	-
Total annual cost, mean ± SD	$1,525 ± $2,467	$262 ± $611	$1,263	<0.001 *
Total medical cost	$1,356 ± $2,326	$214 ± $552	$1,142	<0.001 *
Inpatient	$412 ± $1,135	$33 ± $193	$379	<0.001 *
Outpatient	$663 ± $1,064	$137 ± $351	$526	<0.001 *
ER	$56 ± $218	$36 ± $206	$21	<0.001 *
Home health agency	$203 ± $1,408	$3 ± $63	$201	<0.001 *
Other	$22 ± $116	$6 ± $159	$16	<0.001 *
Total pharmacy cost	$168 ± $404	$48 ± $155	$120	<0.001 *
Age 5 to <13				
Patients	N = 2,126	N = 8,504	-	-
Total annual cost, mean ± SD	$1,227 ± $2,286	$254 ± $582	$973	<0.001 *
Total medical cost	$1,043 ± $2,169	$196 ± $462	$847	<0.001 *
Inpatient	$275 ± $849	$34 ± $214	$240	<0.001 *
Outpatient	$559 ± $1,035	$127 ± $290	$432	<0.001 *
ER	$41 ± $167	$27 ± $124	$14	<0.001 *
Home health agency	$148 ± $1,466	$3 ± $87	$145	<0.001 *
Other	$21 ± $99	$4 ± $31	$17	<0.001 *
Total pharmacy cost	$185 ± $373	$59 ± $262	$126	<0.001 *
Age 13 to <18				
Patients	N = 1,334	N = 5,336	-	-
Total annual cost, mean ± SD	$840 ± $1,351	$304 ± $640	$537	<0.001 *
Total medical cost	$635 ± $1,097	$231 ± $555	$404	<0.001 *
Inpatient	$187 ± $602	$43 ± $275	$143	<0.001 *
Outpatient	$332 ± $596	$149 ± $354	$183	<0.001 *
ER	$36 ± $131	$27 ± $138	$9	<0.001 *
Home health agency	$63 ± $388	$3 ± $51	$61	<0.001 *
Other	$17 ± $73	$9 ± $118	$8	<0.001 *
Total pharmacy cost	$205 ± $529	$73 ± $185	$132	<0.001 *

Notes: **P*-value <0.05
[1] All costs are expressed in 2013 USD.
[2] *P*-values were calculated using Wilcoxon signed-rank tests.

Figure 3. Mean total out-of-pocket annual cost to parents of children with DS and matched controls, by age category



Notes: **P*-value <0.05. [1] *P*-values were calculated using Wilcoxon signed-rank tests to test the null hypothesis of no cost difference between the DS and control groups.

DISCUSSION

- Inpatient out-of-pocket costs during the first year of life were the highest incremental cost incurred by patients with DS, as the need for surgery can be greatest during this time period when about 40-50% of children with DS are identified as having cardiac conditions, gastrointestinal complications, or cataracts that often require surgical treatment[8,11,12]
- Children with DS develop fewer medical conditions requiring hospitalization as they age, resulting in decreased out-of-pocket inpatient costs to parents
- Incremental out-of-pocket costs for outpatient visits, emergency room visits, home health agencies, and pharmacy costs were greater for patients with DS than their matched controls, but did not vary to a great extent across age groups
- The costs estimated represent costs to parents of children with DS while other researchers, like Cuckle et. al, estimated lifetime incremental costs that included direct and indirect costs; it is difficult to compare results from this study with results from other studies that do not estimate individual out-of-pocket costs

LIMITATIONS

- As administrative claims data were used to estimate costs, missing information may have resulted in selection bias, confounding, and measurement error
- Small differences between the DS case group and control group remained after matching for certain age cohorts, though these differences are unlikely to strongly bias results
- This study focuses only on out-of-pocket health care costs to parents of children with DS, which is only one of many costs associated with raising a child that parents must consider; the study does not consider costs of educational, developmental, or other services children with DS may need
- In addition, non-economic factors such as emotional considerations, values, demands on time, or other hardships parents of children with DS may face are not examined in this study, though these factors are often considered to be as important as economic issues parents face
- Incremental lifetime costs can be difficult to measure due to lack of data and complexities in different aspects of life over the life course of a child with DS

CONCLUSION

- Parents of children with DS pay an additional $84 per month in out-of-pocket medical expenses, on average, when costs are amortized over 18 years
- Mean total out-of-pocket medical costs are higher for parents of children with DS across all age categories
- Efforts are being made to establish a population-based national registry for people with DS[13], which may include data on daily costs incurred by a person with DS and thus allow for the calculation of more accurate out-of-pocket expenses
- This study addresses only out-of-pocket medical expenses to parents associated with raising a child with DS; other non-financial costs and considerations related to emotional impact, personal values, the need for assistance, and time demands that many consider equally as important as financial costs are not explored in this study
- Expectant parents are faced with a with a large number of factors to consider when making pregnancy decisions; out-of-pocket medical costs are only one aspect that figure into this decision when parents receive a prenatal diagnosis of DS

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