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MANAGED CARE

Economic Benefits of Increased Diagnosis of Celiac Disease in a National Managed Care Population in the United States

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Objectives.—To estimate the rate of celiac disease diagnosis and evaluate the economic benefits of diagnosis by analyzing retrospective cohorts from a national managed-care-population database.

Methods.—We identified patients who received a new diagnosis of celiac disease. We also identified 3 control groups, persons without a diagnosis of celiac disease but who exhibited 1, 2, or 3 or more symptoms associated with the disease. Using claims, encounter, and eligibility data of ~10.2 million managed care members across the United States between January 1999 and December 2003, we measured and compared direct standardized relative value based (RVU) medical costs and utilization of selected health care services among the 4 study cohorts.

Results.—The rate of new diagnosis for celiac disease more than doubled over the 4-year period. The celiac disease cohort had a significant trend reduction in direct standardized medical costs relative to the three control groups. RVU-based medical costs in the celiac cohort were 24%, 33%, and 27% lower than cohort 1 ($p<0.05$), 29.0%, 38%, and 24% lower than cohort 2 ($p<0.05$), and 38%, 33%, and 31% lower than cohort 3 ($p<0.01$) for the 12-month, 24-month and 36-month post-diagnosis periods, respectively. The reductions in costs were attributable to decreasing trends in utilization of office visits, lab, diagnostic, imaging, and endoscopy procedures relative to the 3 comparative cohorts over the 3-year follow-up period.

Conclusions.—There was an increase in the rate of celiac disease diagnosis, which was associated with significant reduction in direct standardized RVU-based medical costs and utilization of selected health care services over time.

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Celiac disease is an autoimmune disorder, that occurs in genetically predisposed individuals due to an immune response to gluten,[1] the storage protein of wheat. Those with celiac disease also react to similar proteins in rye and barley. Celiac disease is considered to affect about 1% of the population.[2–5] The majority of those with the disease are undiagnosed. The clinical presentation of celiac disease is tremendous-

ly variable. Patients can present critically ill with diarrhea and a classical malabsorption syndrome, or with atypical symptoms that can affect any organ system.[6] The atypical, non-diarrheal presentations are now the most frequent.[7] It is this diverse mode of presentations that make the diagnosis difficult to make.

While the rate of diagnosis is increasing, the clinical prevalence does not approach the actual prevalence of the disease based on antibody screening,[8,9] indicating that celiac disease is grossly under-diagnosed in the United States. Under-diagnosis may be attributed to lack of awareness of the wide clinical spectrum of the disease as well as under-use of serological tests by physicians.[10] The average delay in diagnosis for adult patients with celiac disease ranges from 4 to 11 years in North America.[7,11,12] Under-diagnosis and delay in the disease is associated with a high rate of patient dissatisfaction.[11]

It is generally agreed that patients with celiac disease should be identified and treated, regardless of whether the patients have the typical symptoms or an associated condition. Diagnosis and treatment with a gluten-free diet leads to improvement in quality of life, even in those asymptomatic at diagnosis.[11,13]

In view of the high rate of under-diagnosis of celiac disease, we used the data from a large national managed-care population to evaluate whether there has been an increase in the rate of celiac disease diagnosis and an associated reduction in health care expenditure.

METHODS

Data Source

CIGNA HealthCare has approximately 10.2 million enrolled members across the United States. Their claims, encounter and eligibility data from 1999 to 2003 served as the data source for this study. All medical claims except laboratory claims were used to determine celiac disease.

Diagnosis

The ICD-9 code 579.0 constitutes: celiac crisis, infantilism, rickets, Gee-Herter disease, gluten enteropathy, idiopathic steatorrhea, nontropical sprue. This diagnosis code was used as a surrogate for a clinical diagnosis of celiac disease, as we had no opportunity to review charts in order to confirm the diagnosis. Members who received a primary or secondary diagnosis of 579.0 in any settings were considered to be celiac disease cases. We used the rate of coding as a surrogate for the rate of diagnosis of celiac disease.

The cumulative annual rate for celiac disease diagnosis was calculated as the number of new diagnosis of celiac disease over the 12-month time period divided by the number of member years at risk during the same time period. A direct standardization method was used to calculate age- and sex-adjusted rate of new diagnosis using 2000 as the standard population.

Study Groups

A retrospective cohort study design was used to evaluate the economic benefits and cost-savings associated with diagnosis of celiac disease. Eligible members were cohort members who were 62 years or younger at the time of diagnosis, were continuously enrolled with CIGNA HealthCare without a gap during the 12-month time period prior to diagnosis, and were not eligible for Medicare during the 3-year follow-up period.

All 525 members who received an initial primary or secondary diagnosis of celiac disease in 2000 and met the eligibility criteria were assigned into the celiac disease cohort. We additionally constructed 3 controls groups, based on persons who did not have a diagnosis of celiac disease. Over 2.7 million members exhibited at least one systemic, gastrointestinal or nutritional manifestations of symptoms associated with celiac disease in 2000. Members were stratified into 3 mutually exclusive groups of members who

exhibited 1, 2, or 3 or more symptoms associated with celiac disease (but who did not have a diagnosis of celiac disease). A 2-stage random sampling process was used to select members to be included in the study. In the initial stage, 50,000 members were randomly selected from each cohort to check for eligibility and a negative history of celiac disease diagnosis. Subsequently, a random sample of 1200 members was drawn from each of the three samples. Members who were younger than 62 years of age and were eligible anytime for Medicare were further dropped from the sample, resulting in 1109, 1038, and 980 members in each of the 3 comparative cohorts who exhibited 1, 2, or 3 or more symptoms associated with celiac disease, respectively.

Study Outcomes

We utilized a standardized unit cost in arriving at medical costs because variations in billed charges or actual payment were subject to regional or geographical differences in wage, price, or negotiated contract terms and may not truly reflect differences in health care needs or resource consumptions. We used the Resource-Based Relative Value Scale (RBRVS) approach developed by the Medicare Prospective Payment Commission.[14,15] Relative Value Units (RVUs) are assigned by Center for Medicare and Medicaid Services (CMS) to physician service to reflect level of physician work, practice expenses, and malpractice costs for the service [Total RVU = Work + Practice Expense (PE) + Malpractice (PLI)].[16] Cost in 2003 dollars was calculated as number of RVU units multiplied by 2003 national conversion factor of 36.7856. For services not included in the physician fee schedule (eg, laboratory services etc), 2003 national fee schedule files available from CMS's Web site were used to calculate standardized price for the service.[16] The medical claims for inpatient admissions and services where RVU information were not available were not included. Moreover, costs and utilization related to outpatient pharmacy and mental health claims were excluded because a substantial percentage of enrolled members do not have pharmacy and mental health benefits. Thus, RVU-based medical costs reflects costs for portion of medical services pertaining to physician services, imaging, laboratory services, durable medical equipment, and parenteral and enteral nutritional services. Partial-year costs for members who were not continuously enrolled for any 12-month study period were annualized.

Berenson-Eggers type of service classification was used to classify type of services based on CPT procedure codes into physician office visits, lab and diagnostic tests, imaging, and endoscopy procedures. Emergency room visits were classified based on a combination of Berenson-Eggers classification, revenue codes, and CIGNA-specific codes. Rate of utilization of selected services and hospital admissions for the 4 study cohorts during a given 12-month time period were calculated.

Statistical Analyses

Statistical analysis was performed using SAS version 8.2. Analysis of variance and chi-square statistic was used to determine whether age, risk score, and the distribution of patient's baseline demographic characteristics, eg, gender, region, and benefit structure, differed between the celiac disease cohort and the 3 comparative cohorts. Primary or secondary diagnosis codes during office visits, ambulatory visits, ER visits, and inpatient visits during the 12-month period prior to the index diagnosis date were used to classify patients' comorbidities into 259 clinically homogenous groups.[17] Comorbid conditions that were prevalent in at least 0.5% of patients were used in developing risk scores to account for differences in patient's baseline risk.

The study employed a difference-in-difference analysis that used trend or change in the level of outcome in each of the 3 comparative cohorts as a control for what would have

happened in the absence of celiac diagnosis in the celiac disease cohort. The methodology has been used in various evaluative and economic studies to determine the impact of specified intervention, program, or policy changes on outcomes.[18–21]

The model parameters of interest are the difference-in-difference parameters that reflect the interaction effects between study cohorts and time period of measurement. The presence of a significant interaction effect indicated that the mean change in the study outcome in a given time period from the baseline period in the celiac disease cohort is significantly different from the mean change in the study outcome between the same time periods in the comparative cohort.

RESULTS

The age- and sex-adjusted rate of new diagnosis for celiac disease more than doubled over the 4-year period of our study (Figure 1). This was higher in females than in males, and increased with age (not shown). The value was 265 per million members per year in females and 145 per million members per year in males in 2003. The rate of new acquisition of this diagnostic code was highest among members aged 65 years and older at 343 per million members per year and lowest among members between 18–29 years at 153 per million members per year in 2003. There were, however, increases in all age groups (Table 1).

To assess cost savings, we created cohorts of individuals with diseases or symptoms associated with celiac disease in 2000 and who were continuously enrolled without gaps. We also evaluated the association of celiac disease diagnosis over time among these cohorts (Table 2). We found the higher the number of associated conditions exhibited by members, the higher the likelihood of celiac disease diagnosis over the 4-year study period. For those exhibiting any one of the associated conditions, celiac disease diagnosis occurred

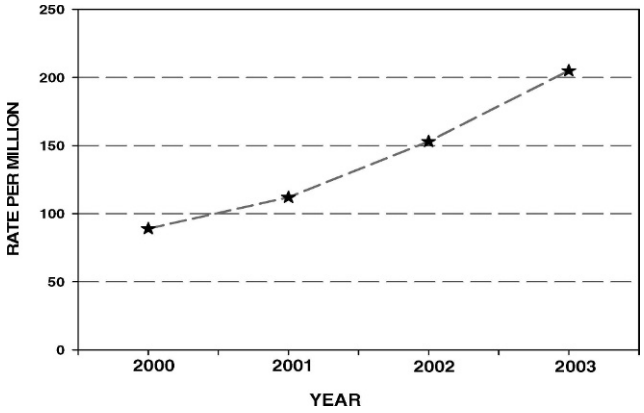

Figure 1. *Age and Sex Standardized Rate of Celiac Disease Diagnosis among CIGNA HealthCare Managed Care Population from 2000–2003.*

in 0.99 per 1000, compared to 4.03 per 1000 in those exhibiting 3 or more associated conditions. The top 5 conditions with the highest occurrence of celiac disease diagnosis in the study period were dermatitis herpetiformis (1 in 34), followed by Down syndrome (1 in 53), nutritional marasmus (1 in 65), cachexia (1 in 98), and pancreatic insufficiency (1 in 123).

The demographic and baseline characteristics of members in the study cohorts at the time of diagnosis are shown in Table 3. Members of the celiac disease cohort were similar in mean age to cohort 1 and were significantly younger than cohorts 2 or 3. The celiac disease cohort had a higher risk score for medical costs than cohort 1 and a lower risk score than cohort 2 and 3. The celiac disease cohort was less likely to be female than cohorts 2 of the 3 and more likely to be in Mid-Atlantic, Northeast, Tri-Mid, and West Coast regions, and less likely to be in the Southeast and Southwest regions.

Changes in study outcomes in the celiac disease cohort relative to the 3 comparative cohorts using the difference-in-difference model estimates and 95% confidence interval are shown in Table 4. The celiac disease cohort had a significantly decreased trend in RVU-based medical costs relative to the trends observed in the 3 comparative cohorts (Figure 2). RVU-based medical costs in the celiac disease cohort were 24%, 33%, and

Table 1. Rate of Celiac Disease Diagnosis by Gender and Age Group from 2000 to 2003

Year		2000		2001		2002		2003	
		Number[a]	Rate per Million[b]	Number[a]	Rate per Million[b]	Number[a]	Rate per Million[b]	Number[a]	Rate per Million[b]
Gender	Female	661	116	882	145	1187	194	1452	265
	Male	351	62	482	80	664	111	775	145
Age	0–17	281	89	316	96	442	135	508	173
	18–29	95	49	178	87	232	123	253	153
	30–39	155	74	202	92	303	141	376	196
	40–49	209	101	297	135	352	158	458	228
	50–59	163	112	214	134	326	195	368	250
	60–64	61	157	96	215	111	226	139	307
	65+	48	192	61	181	85	213	125	343
	Total	1012	89	1364	113	1851	153	2227	206

[a] Number of enrolled members newly diagnosed with celiac disease in any settings in the time period.
[b] Rate per million member years at risk for the disease during the time.

27% lower than cohort 1, 29%, 37%, and 24% lower than cohort 2, and 38%, 33%, and 32% lower than cohort 3 across the 3-year periods, respectively.

The reduction trend in RVU-based medical costs was attributable to decreasing trends in utilization of office visits, laboratory and diagnostic testing, imaging, and endoscopy procedures. Risk-adjusted rate of office visits in the celiac disease cohort was 13%, 17%, and 12% lower than cohort 1, 19%, 18%, and 8% lower than cohort 2, 21%, 19%, and 15% lower than cohort 3 (Figure 3). Rates of laboratory and diagnostic testing procedures in the celiac disease cohort were 33%, 36%, and 38% lower than cohort 1, 38%, 33%, and 33% lower than cohort 2, and 39%, 33%, and 36% lower than cohort 3 across the same time periods, respectively (Figure 4). The risk-adjusted rates of imaging procedures in the celiac disease cohort were 46%, 35%, and 37% lower than cohort 1, 47%, 29%, and 31% lower than cohort 2, and 48%, 28%, and 30% lower than cohort 3. The reduction in rates of endoscopy procedures was more pronounced during the second- and third-year follow-up period. Rates of endoscopy procedures in the celiac disease cohort were 70%, 78%, and 59% lower in the 24-month, and 60%, 54%, and 32% lower in the 36-month across the 3 cohorts. However, there were no significant differences in mean changes in rates of ER visits and hospitalizations between the celiac disease cohort and the three comparative cohorts.

To determine if there were cost savings for those diagnosed with celiac disease, we assessed the direct medical charges per member per year in 2003 dollars by type of service and overall for the 4-year study period among study members who were newly diagnosed with celiac disease in 2000 (Table 5). The mix of medical services received by members with celiac and direct medical expenditures changed over the 4-year period. Prior to diagnosis, medical charge per member per year was $8502 in 2003 dollars. About one third (30.9%) of the

Table 2. Celiac Disease Diagnosis among Members with Conditions Associated with Celiac Disease in 2000 by Number and Type of Conditions

Number of associated conditions	Numerator[a]	Denominator[b]	Rate per 1,000
Exhibited Any 1	472	477,698	1
Exhibited Any 2	233	106,106	2
Exhibited Any 3 or More	116	28,818	4
Total	821	612,622	1
Condition[c]			
Dermatitis Herpetiformis	6	204	30
Down's Syndrome	11	584	19
Nutritional Marasmus	1	65	15
Cachexia	2	195	10
Pancreatic Insufficiency	2	245	8
Vitamin B Complex Deficiency	11	1600	7
Diarrhea	126	23,775	5
Anemia	84	16,750	5
Crohn's Disease/Regional Enteritis	15	3088	5
Irritable Bowel Syndrome	91	20,169	5
Sjogren's Disease	4	994	4
Weight Loss	2	525	4
Hypocalcemia	2	561	4
Lactose Insufficiency	7	2107	3
Type I Diabetes	61	19,553	3
Osteoporosis	66	21,624	3
Abdominal Pain	296	173,480	2
Fatigue	120	87,945	1
Constipation	29	21,384	1
Enlargement of Peripheral Lymph Nodes	18	14,227	1
Asthma	117	99,150	1
Arthritis	100	86,731	1
Joint Pain	172	181,129	1
Infertility	7	7713	1
Short Stature	0	39	0.00

[a] Number of members in the denominator with a diagnosis of celiac anytime between January 2000 to December 2003.

[b] Number of members with associated conditions of celiac in 2000 who were continuously enrolled without gap from 2000 to 2003.

[c] Members may exhibit more than 1 condition associated with celiac.

expenditures was accounted for by professional services, followed by other medical services (22%), facility inpatient (28%), lab/diagnostic testing (13%), and imaging (8%). At time of diagnosis and 12 months after, facility inpatient expenditures increased almost two fold from $1844 (22%) to $3373 (28%) per member per year. Subsequently, the percentage of direct medical expenditures accounted for by facility services decreased to 15% and 17% during 24 months and 36 months after diagnosis, respectively. Emergency room services made up 5% of expenditures in the 12 months pre-diagnosis, and declined to around 2%–3% of expenditures over the 3-year periods. In contrast, the percent of direct medical costs accounted for by professional and other medical services increased over time from 53% in the 12-month pre-diagnosis period to 59% in the 36-month post-diagnosis period. Overall, the mean medical expenditures per member per year increased from $8502 in the 12-month pre-diagnosis period to $12,024 at time of

Table 3. Demographic and Baseline Characteristics of Study Cohorts at Time of Diagnosis

	Celiac Cohort	Cohort with 1 associated conditions	Cohort with 2 associated conditions	Cohort with 3 or more associated conditions
Number of members	525	1109	1038	980
Mean (SD) age at diagnosis date[a]	33 (19)	35(17)	40(15)	45(12)
Mean (SD) risk score for expected medical costs[b]	8(1)	7(1)	8(1)	9(1)
Gender:[c] Percent male	37	45	37	25
Region[c] (percentages by column)				
Mid-Atlantic region	20	16	18	20
Northeast region	10	4	6	5
Southeast region	10	24	22	22
Southwest region	16	22	25	23
Tri-Mid region	32	24	21	23
West-coast region	12	10	8	7

[a] Mean Difference between CD cohort and cohorts 2 and 3 statistically significant at 0.001 level.
[b] Mean Difference between CD cohort and other cohorts statistically significant at 0.001 level.
[c] Significantly different distribution among the 4 study cohorts at 0.001 level.

Table 4. Difference-in-Difference Risk Ratio (95% Confidence Interval) of Study Outcomes in the Celiac Cohort Relative to the Three Comparative Cohorts during the 4-Year Study Period

Outcomes	Cohorts	12-month post/ pre-	24-month post/pre-	36-month post/pre-
RVU-Based Medical Costs	Celiac / Cohort 1	0.76*** (0.62–0.93)	0.67***(0.52–0.86)	0.73**(0.56–0.95)
	Celiac / Cohort 2	0.71**** (0.60–0.85)	0.62**** (0.50–0.78)	0.75** (0.59–0.96)
	Celiac / Cohort 3	0.62**** (0.52–0.74)	0.67**** (0.54–0.83)	0.68***(0.53–0.87)
Office Visits	Celiac / Cohort 1	0.87*** (0.79–0.96)	0.83*** (0.74–0.95)	0.88* (0.76–1.02)
	Celiac / Cohort 2	0.81**** (0.74–0.89)	0.82*** (0.73–0.93)	0.92 (0.80–1.06)
	Celiac / Cohort 3	0.79**** (0.72–0.86)	0.81**** (0.72–0.91)	0.85** (0.74–0.98)
ER Visits	Celiac / Cohort 1	0.43**** (0.30–0.61)	0.85 (0.57–1.29)	0.92 (0.58–1.46)
	Celiac / Cohort 2	0.44****(0.31–0.62)	1.00 (0.67–1.49)	0.75 (0.49–1.15)
	Celiac / Cohort 3	0.43**** (0.31–0.60)	0.89 (0.61–1.31)	0.76 (0.50–1.16)
Admissions	Celiac / Cohort 1	0.66* (0.41–1.05)	0.94 (0.51–1.73)	0.66 (0.33–1.31)
	Celiac / Cohort 2	1.06 (0.68–1.64)	0.98 (0.55–1.77)	0.74 (0.39–1.41)
	Celiac / Cohort 3	0.76 (0.49–1.16)	0.83 (0.47–1.48)	0.80 (0.42–1.51)
Lab & Diagnostic Testing	Celiac / Cohort 1	0.67**** (0.56–0.79)	0.64**** (0.52–0.78)	0.62**** (0.49–.79)
	Celiac / Cohort 2	0.62**** (0.53–0.72)	0.67**** (0.55–0.81)	0.67**** (0.53–0.84)
	Celiac / Cohort 3	0.61**** (0.52–0.72)	0.67**** (0.55–0.80)	0.64**** (0.51–0.80)
Imaging	Celiac / Cohort 1	0.54**** (0.43–0.67)	0.65**** (0.51–0.83)	0.63**** (0.48–0.82)
	Celiac / Cohort 2	0.53**** (0.44–0.65)	0.71*** 0.57–0.88)	0.69*** (0.54–0.87)
	Celiac / Cohort 3	0.52**** (0.43–0.63)	0.72*** (0.59–0.88)	0.70*** (0.55–0.87)
Endoscopy	Celiac / Cohort 1	1.17 (0.82–1.66)	0.30**** (0.19–0.49)	0.40**** (0.25–0.64)
	Celiac / Cohort 2	0.71** (0.52–0.96)	0.22**** (0.14–0.35)	0.45**** (0.28–0.71)
	Celiac / Cohort 3	0.85 (0.64–1.12)	0.41**** (0.27–0.61)	0.68* (0.45–1.04)

* $p<0.1$; ** $p<0.05$; *** $p<0.01$; **** $p<0.001$.

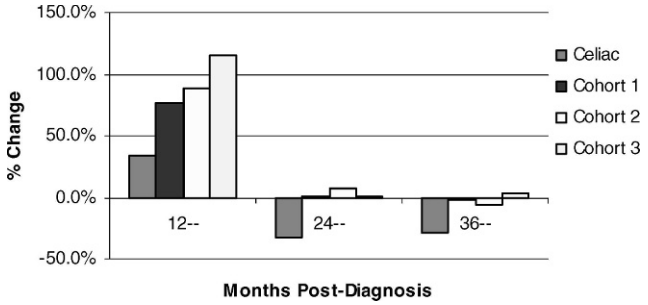

Figure 2. *Percent Change in Average RVU-based Medical Cost from Pre-Diagnosis Period.*

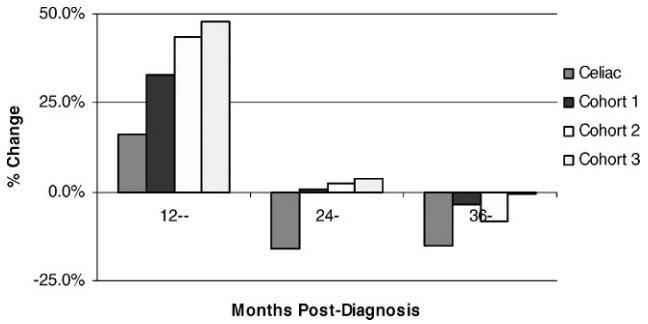

Figure 3. *Percent Change in Rate of Office Visits from Pre-Diagnosis Period.*

diagnosis and 12-month post and subsequently decreased to $7133 and $7854 for the 24-month and 36-month period after clinical diagnosis of celiac, respectively.

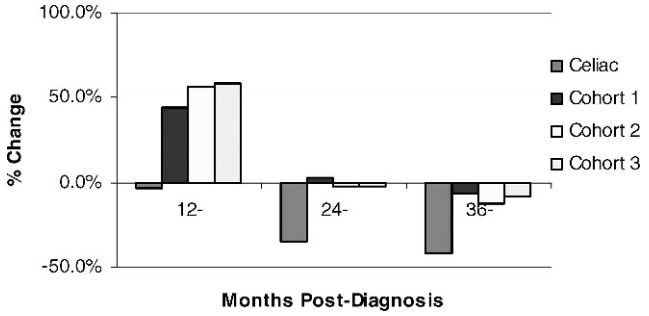

Figure 4. *Percent Change in Rate of Lab and Diagnostic Testing from Pre-Diagnosis Period.*

DISCUSSION

This study, involving over 10 million subjects enrolled in managed care programs, from all regions of the United States, reported that the age- and sex-adjusted rate of new diagnosis for celiac disease increased over 2-fold from 2000 to 2003.

This increasing rate of diagnosis, assuming diagnosis of celiac disease by the 579.0 code, is in line with that reported in another population based study from Olmsted County, Minnesota where Murray et al reported the annual incidence of celiac disease increased from 0.9 per 100,000 in 1950–1989 to 3.3 per 100,000 in the 1990s. In the last 2 years of that study (2000 and 2001), the incidence was 9.1 per 100,000.[8] Taking the increase in

Table 5. Medical Charges per Member per Year (PMPY) with diagnosis of Celiac Disease in 2003 Dollars and % of Medical Charges by Type of Services over the 4-Year Study Period

	12-month pre- (n=525 member years)		12-month post- (n=447 member years)		24-month post- (n=327 member years)		36-month post- (n=245 member years)	
Service Type	Billed Charge PMPY	% Charges	Billed Charge PMPY	% Charges	Billed Charge PMPY	% Charges	Billed Charge PMPY	% Charges
ER	$388	6%	$282	2%	$203	3%	$213	3%
Facility Inpatient	$1844	22%	$3373	28%	$1091	15%	$1328	17%
Imaging	$660	8%	$712	6%	$621	9%	$570	7%
Lab/Diagnostic Testing	$1102	13%	$1155	10%	$923	13%	$1085	14%
Professional Services	$2625	31%	$3454	29%	$2172	30.%	$2590	33%
Other Medical Services	$1884	23%	$3048	25%	$2122	30%	$2067	26%
Total	$8502	100%	$12,024	100%	$7133	100%	$7854	100%

the diagnosis of celiac disease in our database, the crude rate calculated in our study was 8.9 per 100,000 in 2000, similar to the Olmsted County level. This value increased to 20.6 per 100,000 in 2003. The findings that the increase in diagnosis of celiac disease was higher in females than in males and increased with age is consistent with those reported in the literature.[8,22] In addition an increase was noted in all age groups.

However, our findings of an increase in diagnosis of celiac disease may be due to an increased consciousness of celiac disease or due to physician behavior as explained by the well known health economic concept of supplier induced demand. Although clinically manifest cases were detected with the 579.0 diagnosis, we may have missed data points on patients with clinically silent and atypical presentations. Alternatively, the diagnosis may be underestimated, as not all patients with celiac disease may seek care during the study time period and will not have relevant claims in the database.

In this study we explored the economic benefit of celiac disease diagnosis. In order to explore this cohort groups were constructed with increasing numbers of symptoms or diseases associated with celiac disease. These cohorts did in fact have increasing rates of celiac disease diagnosis, confirmation of the value of using these cohort groups. We found members in the celiac disease cohort had a significant reduction in RVU-based direct medical costs when compared with control cohort members. The reduction in outcome trends observed in the celiac cohort was observed consistently across the 3 comparative cohorts varying in number of conditions associated with celiac disease and was due to decreasing trends in utilization of office visits, laboratory and diagnostic testing, imaging, and endoscopy procedures.

The overall savings after celiac disease diagnosis was confirmed when we examined the mean direct medical expenditures per capita among members who received the diagnosis for the celiac disease diagnosis. These expenditures increased substantially at the time of diagnosis and for the 12-months after the diagnosis of celiac disease, when compared to 12-month prior to diagnosis. The increase in expenditures was attributed mainly to an increase in the facility inpatient component. However, over time, the mean medical expenditures decreased from $8502 per capita to $7133 and $7854 for 24-month and 36-month after diagnosis. This was due to a decrease in facility inpatient and emergency room expenditures and an increase in professional services and other medical services expenditures over the 4-year period. This trend of increased expenditure in the immediate post diagnosis period is consistent with clinical practice. Patients often after diagnosis get screened for co-morbid conditions such as vitamin and mineral deficiencies, anemia and osteoporosis. They often undergo inappropriate screening for malignancies as well.

The reliability of our data and methods is attested by the fact that a significant number of the cohorts with specific associated diagnoses were in fact diagnosed with celiac disease over the 4-year study period. This was seen in patients with dermatitis herpetiformis where only 2.9% received a diagnosis for celiac disease; although 100% patients with this diagnosis actually do have celiac disease, the majority evidently did not receive the 579.0 diagnosis label. The failure to identify members with dermatitis herpetiformis may be due to the fact that while all subjects with the disease have gluten sensitivity, not all manifest gastrointestinal symptoms. In addition, dermatologists do not always recommend a gluten-free diet, and the celiac disease diagnosis may not be coded for unless there is referral and evaluation by a gastroenterologist. Similarly, those with the diagnosis of Down syndrome had a celiac disease diagnosis rate of 2% comparably lower than the reported incidence rate of 6.3%.[23] These results support the concept that while there is an increasing rate of celiac disease, it remains under

diagnosed in the United States, even in groups known to be at increased risk.

Although, a fraction of the comparison groups may actually have celiac disease, we observed significant trend reduction in costs and utilization over time in the celiac cohort relative to the comparison groups. Moreover, the higher the likelihood of members having the celiac disease diagnosed in the later years, the higher the magnitude of the differences in the cost trend. The observed cost difference was highest in cohort 3, relative to cohorts 1–2. The findings seem to indicate that if we were able to limit the comparative cohort to only members who were eventually diagnosed with celiac disease later on, the observed cost-difference would have been greater. Therefore, the study may have underestimated the cost savings or differences in costs associated with detection of celiac disease over time.

Limitations of the study should be noted. The study is observational in nature, and members were not randomly assigned to treatment groups. The study controlled for differences in age, gender, benefits structure, geographical region, and presence of comorbid conditions prior to diagnosis. Nevertheless, other variables not considered in the study may affect the results. Secondly, the study excluded mental health and pharmacy claims because substantial percentages of members do not have coverage for the services. Thirdly, the study relied on administrative claims data as data sources. Administrative data are created to support the payment and other administrative functions of health plans. Detailed clinical data elements are not available, and quality of diagnosis may vary among providers. This may lead to false identification of diseased members (over diagnosis) or failure to identify members with the disease (under diagnosis). Despite the disadvantages, claims data offered important advantages in health services research. Administrative data are readily available and provide an economical approach to studying and investigating costs and utilization of medical services over time. Using these data we demonstrated economic benefit with the diagnosis of celiac disease.

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