

Development of a new comorbidity index for adults with cerebral palsy and comparative assessment with common comorbidity indices

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ABBREVIATIONS

AIC	Akaike Information Criterion
CCI	Charlson Comorbidity Index
ECI	Elixhauser Comorbidity Index
WCI	Whitney Comorbidity Index

AIM To develop a new comorbidity index for adults with cerebral palsy (CP), the Whitney Comorbidity Index (WCI), which includes relevant comorbidities for this population and better predicts mortality than the Charlson Comorbidity Index (CCI) and Elixhauser Comorbidity Index (ECI).

METHOD Data from the Optum Clinformatics Data Mart was used for this retrospective cohort study. Diagnosis codes were used to identify adults aged 18 years or older with CP ($n=1511$ females, $n=1511$ males; mean [SD; range] age=48y [19y 2mo; 18–89y]) and all comorbidities in the year 2014. The WCI was developed based on the comorbidities of the CCI and ECI and other relevant comorbidities associated with 2-year mortality using Cox regression and competing risk analysis. The WCI was examined as unweighted (WCI_{unw}) and weighted (WCI_w). The model fit and discrimination (C-statistic) of each index was assessed using Cox regression.

RESULTS Twenty-seven comorbidities were included in the WCI; seven new comorbidities that were not part of the CCI or ECI were added. The WCI_{unw} and WCI_w showed a better model fit and discrimination for 1- and 2-year mortality compared to the CCI and ECI. The WCI_{unw} and WCI_w were strong predictors for 1- and 2-year mortality (C-statistic [95% confidence interval] ranging from 0.81 [0.76–0.85] to 0.88 [0.82–0.94]).

INTERPRETATION The new WCI, designed to include clinically relevant comorbidities, provides a better model fit and discrimination of mortality for adults with CP.

The transition from childhood to adulthood for individuals with cerebral palsy (CP) is accompanied by an early and elevated burden of disease across biological systems and mental health disorders.^{1–15} Outcome-based research regarding aging with CP throughout the adult lifespan is in its infancy and there is a critical need to identify appropriate methods for risk adjustment that can also be used for clinical monitoring.

In most outcome-based research, risk adjustment for patient demographics and health status (among other factors) is necessary to obtain clinically meaningful results. Among the risk adjustment methodologies, utilizing comorbidity indices is a widely popular method that serves as a proxy for health status. The two most commonly used validated measures are the Charlson Comorbidity Index (CCI), which includes 17 weighted comorbidities,^{16,17} and the Elixhauser Comorbidity Index (ECI), which includes 30 unweighted comorbidities,¹⁸ which were designed to predict 1-year and in-hospital mortality respectively, among other outcomes in the general population.

However, the CCI and ECI are not a ‘one size fits all’ approach to comorbidity indices and may not be applicable across clinical conditions, especially for CP. The comorbidities from the CCI and ECI were selected from a non-CP sample and may not capture the prevalent comorbidities that may be associated with mortality specific to adults with CP. For example, constipation, neurogenic bowel or bladder, sleep disorders, and dysphagia are common problems for individuals with CP^{19–21} that can impact health and survival, but are not in the CCI or ECI. Emerging evidence suggests that bone fragility (e.g. fragility fracture) may be involved in the pathogenesis of unhealthful aging, possibly by triggering or exacerbating physiological decline of the cardiorespiratory system.^{22,23} However, markers of or associated with bone fragility, such as osteoporosis, fractures, and osteoarthritis, are not part of the CCI or ECI.

The lack of a CP-specific comorbidity index has implications for research in terms of adequately addressing confounding by health and disease status for clinical discoveries. Furthermore, some electronic medical record

systems (e.g. Michigan Medicine Health System) automatically populate the CCI and/or ECI based on user input to assist in clinical decision-making. Therefore, there are various needs for research and clinical practice to develop a comorbidity index specific to the unique comorbidity profiles of adults with CP. Accordingly, the purpose of this study was to develop a new comorbidity index, termed the Whitney Comorbidity Index (WCI), which predicts mortality better than the CCI and ECI by accounting for clinically relevant comorbidities specific to adults with CP. The hypothesis was that the newly developed WCI would have better discrimination and model fit for mortality among adults with CP compared to the CCI and ECI.

METHOD

Data source

Data from full calendar years 2014 to 2016 were ascertained from the Optum Clinformatics Data Mart database (OptumInsight, Eden Prairie, MN, USA), which is a nationwide, deidentified single private payer administrative claims database, as described previously in detail.²⁴ Briefly, to be enrolled in private insurance, the individual either pays for their insurance or is covered by their employer or their spouse's employer. For these reasons, this database may reflect a slightly more affluent sector of the population and less severely affected sector of the population with CP. Administrative claims data are used primarily for billing reimbursement of healthcare visits and health conditions are identified using specific codes (discussed later in the article) attached to individual claims. Since data are deidentified, the University of Michigan institutional review board approved this study as non-regulated.

All medical conditions were identified using the International Classification of Diseases, Ninth Revision, Clinical Modification (ICD-9-CM)²⁵ codes given the time frame of the study and use of ICD code versions in the USA. See Table S1 (online supporting information) for a list of the ICD-9-CM codes used to identify comorbidities in the WCI. The ICD-9-CM codes for CCI and ECI comorbidities have been published elsewhere.^{16–18}

Participant selection

The calendar year 2014 was used to identify eligible participants: adults aged 18 years or older with one or more claims for CP on two or more separate days to enhance specificity; continuous enrollment in a health plan; and with two or more health service utilizations to limit detection bias. The requirement for unbroken health plan enrollment in 2014 was to ascertain baseline comorbidity data, as described later in the article. Participants were then followed from the 1st January 2015 to death, loss to follow-up, or end of the study period (31st December 2017), whichever came first.

Data about the severity of CP using common clinical measures (e.g. Gross Motor Function Classification System) are not available in insurance claims data. Furthermore, more than 70% of the CP sample had 'other' or

What this paper adds

- Common comorbidity indices exclude relevant comorbidities for adults with cerebral palsy (CP).
- A new comorbidity index for adults with CP was created by harmonizing clinical theory and data-driven methods.
- The Whitney Comorbidity Index better predicted 1- and 2-year mortality than other commonly used comorbidity indices.

'unspecified' CP.⁶ Therefore, the clinical subtypes of CP (e.g. hemiplegic, athetoid) could not be accounted for. Based on previous studies,^{1,2,6,26} this privately insured sample of adults with CP may reflect a slightly less severely affected segment of the population with CP.

Outcome

All-cause mortality was determined as the number of days from the 1st January 2015 to 31st December 2017 (2-year period). Date of death was derived from the Date of Death table view in the database. This table is kept current and is sourced from the Death Master File, which is maintained by the Social Security Office.

CCI and ECI

This study used modified versions of the CCI and ECI. The original CCI,¹⁶ which was adapted by Deyo et al.¹⁷ for use with ICD-9-CM administrative claims data, includes 17 weighted comorbidities. The original ECI includes a score (yes/no) for 30 comorbidities and is unweighted.¹⁸ Due to overlap with CP, this study omitted hemiplegia/paraplegia from the CCI and paralysis from the ECI, resulting in a total of 16 weighted comorbidities for the modified CCI and 29 unweighted comorbidities for the modified ECI (hereafter referred to as CCI and ECI).

WCI

Development of the WCI involved an iterative process that attempted to harmonize clinical theory and existing validated measures (i.e. CCI, ECI) using data-driven approaches. Informed by the literature, a comprehensive list of comorbidities present in claims data across all biological systems were initially considered.^{1–15} Comorbidities were excluded if they are known to have poor sensitivity or specificity for claims-based research, such as obesity, or require an adult CP clinical specialist to make an accurate diagnosis for complex conditions that non-CP specialists may not be able to accurately make, such as fatigue.

Of the remaining comorbidities clinically relevant to CP, CCI and/or ECI comorbidities were examined and selected where appropriate (e.g. myocardial infarction from the CCI, hypertension from the ECI). For comorbidities that had overlap in codes (e.g. CCI renal disease and ECI renal failure), the comorbidity with the strongest association with mortality was selected, as determined by Cox proportional hazards regression, which is discussed later. Some comorbidities were combined for simplicity in implementing the WCI (e.g. mild-to-severe liver disease from the CCI, blood loss and deficiency anemias from the

ECI). Other codes for comorbidities were modified to increase relevance to CP (e.g. chronic respiratory failure was added to CCI chronic pulmonary disease). Epilepsy was removed from the 'comorbidity, other neurological disorder' from the ECI to have epilepsy as a standalone comorbidity, given its relevance to CP.

To expand the list, comorbidities that may be associated with health status and mortality among adults with CP were examined. Intellectual disabilities and autism spectrum disorder were added each as their own comorbidity since they can co-occur with CP and can increase the medical complexity of the individual with CP. Pneumonia was added as its own comorbidity because it is a common cause of premature mortality for individuals with CP.²⁷ Other added comorbidities included anxiety, bone fragility (i.e. osteoporosis or fracture), rheumatoid arthritis and other inflammatory polyarthropathies, osteoarthritis and allied disorders, sleep disorders, gastrointestinal issues (i.e. peptic ulcer disease excluding bleeding from the ECI, constipation, inflammatory bowel disease, irritable bowel syndrome), neurogenic bowel or bladder (as one comorbidity), and dysphagia.

Given the importance of ambulation on health outcomes for individuals with CP, a proxy measure for ambulatory status as yes versus no was considered and the methods were guided by Pulgar et al.²⁸ who used a Medicaid sample of children with CP; 25.4% of the study sample could be categorized as non-ambulatory, but 66.5% could not be assigned an ambulatory status. Given the unknown validity, an administrative-based ambulatory status variable was not considered for inclusion in the WCI. Nonetheless, the severity of CP is associated with multi-morbidity⁵⁻¹⁰ and may not add substantial prognostic value as long as relevant comorbidities are included in the index.

In total, 33 comorbidities were initially considered for inclusion in the WCI before exclusion of comorbidities that were not associated with mortality.

Statistical analysis

Descriptive characteristics were summarized for the sample. Cox proportional hazards regression models were developed before (unadjusted) and after adjusting for age, sex, and US region of residence, with mortality up to 2 years as the outcome. Each comorbidity from the CCI, ECI, and initial 33 comorbidities considered for the WCI were included in the model separately as the primary exposure variable to determine the unadjusted and adjusted association with 2-year mortality as a hazard ratio (HR) with 95% confidence intervals. Models were developed with each primary exposure separately (before and after adjusting for age, sex, and US region of residence) rather than all exposure variables in a single model, which would lead to low confidence of the residuals, thus biasing and impacting the validity of the statistical model.²⁹

Of the 33 comorbidities initially considered for the WCI, comorbidities were selected if the unadjusted or adjusted association with mortality had a $p < 0.20$. The p -

value threshold was selected because a more stringent threshold could fail to include important comorbidities. The final set of comorbidities from the WCI was then examined with each comorbidity unweighted and denoted as WCI_{unw} . To determine if weighting the final WCI comorbidities, denoted as WCI_w , would improve the model fit or discrimination for predicting 2-year mortality, the following weighting algorithm was applied, which was guided by the process outlined by Quan et al.³⁰: a weight of 1 for an adjusted HR less than 1.50; a weight of 2 for an adjusted HR between 1.50 and 2.49; a weight of 3 for an adjusted HR between 2.50 and 3.49; a weight of 4 for an adjusted HR between 3.50 and 4.49; a weight of 5 for an adjusted HR between 4.50 and 5.99; and a weight of 6 for an adjusted HR equal to or greater than 6.00.

To determine if the WCI (as WCI_{unw} and WCI_w) is a better measure to predict 2-year mortality compared to the CCI and ECI, Cox proportional hazards regression models were developed. The base model was adjusted for age, sex, and US region of residence. Each comorbidity index was added separately to the base model (as continuous and discrete in separate models) and model fit and discrimination statistics were computed. The Akaike Information Criterion (AIC) was used to assess goodness of fit. The concordance statistic (C-statistic) was used to assess the discrimination of each model (equivalent to the area under the receiver operating characteristic curve). The C-statistic ranges from 0.50 to 1.00 with a value greater than 0.80 indicating a strong model. This study initially computed both the C-statistic proposed by Harrell and Uno due to differences in how censoring is handled.³¹ However, given the similarity of results and conclusions, the Uno C-statistic and 95% confidence intervals are reported. The concordance probabilities were compared between each model using the methodology outlined by Uno et al.³¹ designed for censored survival analysis.

The assumption of proportional hazards was met for all models; it was examined by Kaplan-Meier plots and by creating an interaction of the exposure and a function of survival time in the model.

Sensitivity analysis

Given the potential for loss to follow-up common with longitudinal claims-based studies, unadjusted (Gray's test for equality of cumulative incidence functions) and adjusted (subdistribution hazard model) competing risk analysis was performed when examining the association between each comorbidity from the CCI, ECI, and WCI with 2-year mortality to determine if loss to follow-up was a competing event. If there was no evidence of a competing risk, the findings from the traditional Cox model were used.

Ethnic grouping was not included for statistical adjustment due to the extent of missing or unknown data. We assessed for possible confounding and selection bias, as described previously,⁷ when comparing the AIC and C-statistics between the different comorbidity index

models. Briefly, we restricted the sample to individuals with complete data on ethnic grouping and performed analysis before and after adjusting for ethnic grouping. Evidence of confounding by ethnic grouping was determined by comparing results from the restricted sample before and after adjusting for ethnic grouping. Evidence of selection bias was determined by comparing results from the non-ethnic grouping-adjusted restricted sample to the non-ethnic grouping-adjusted full sample.

Since the CCI and ECI were developed for mortality time intervals of less than 2 years, comparing the model fit and discrimination statistics was performed with 1-year mortality as the outcome.

Analyses were performed using SAS v9.4 (SAS Institute, Cary, NC, USA).

RESULTS

The baseline descriptive characteristics of adults with CP ($n=3092$) are presented in Table 1. During the follow-up for a mean (SD) and median (interquartile range) of 595 (215) and 730 (395–730) days respectively, 73 adults with CP died (2.4%), 1261 were right-censored due to loss of follow-up before the end of the study period (40.8%), and 1758 (56.9%) were right-censored due to the end of the study period.

WCI: individual comorbidities

The prevalence and HR from the traditional Cox model and competing risk model for each comorbidity from the CCI are presented in Table S2 (online supporting information), for each comorbidity from the ECI in Table S3 (online supporting information), and for each comorbidity from the WCI in Table 2. Of the 33 comorbidities initially considered for the WCI, six did not meet the statistical criteria, resulting in a total of 27 comorbidities. Table 3 shows the individual comorbidities included in the final

WCI and any modifications made from the CCI or ECI comorbidities.

Index score comparison

Due to the low number of outcome events, adjusted analysis of metastatic cancer was not possible but exhibited a large HR in the unadjusted model. Therefore, a weight of 6 was given to this comorbidity for the WCI_w to be consistent with the CCI weight.

The mean (SD; range) of the CCI, ECI, WCI_{unw} , and WCI_w score was 1.09 (1.69; 0–14), 2.61 (1.47; 0–15), 3.19 (2.86; 0–17), and 6.35 (6.01; 0–36) respectively. The AIC and C-statistic measures for 1- and 2-year mortality are presented in Table 4. Regardless of whether the WCI was weighted or not, both WCI_{unw} and WCI_w exhibited an improved model fit (i.e. lower AIC) and discrimination (i.e. higher C-statistic values) compared to the CCI and ECI when the indices were examined as continuous and discrete for 2-year mortality. The improved discrimination was significantly higher for the WCI_{unw} and WCI_w compared to the CCI and for the WCI_w compared to the ECI (all $p<0.05$) when the indices were treated as continuous (WCI_{unw} compared to ECI, $p=0.169$) but not when they were treated as discrete (all $p>0.34$). When the outcome was 1-year mortality, the patterns of AIC and C-statistic measures were similar to the main analysis.

In the sample restricted to individuals with complete data on ethnic grouping ($n=2639$), the patterns of AIC and C-statistic measures were similar to the main analysis, with little to no difference between models with and without ethnic grouping adjustment (Table S4, online supporting information). Further, the C-statistic measures were similar between the full sample and ethnic grouping-restricted sample without adjustment for ethnic grouping. These findings do not provide evidence of confounding or selection bias by ethnic grouping.

DISCUSSION

Using a nationwide private insurance database, the WCI was developed by harmonizing clinical theory (i.e. clinically relevant comorbidities for CP) and existing validated comorbidity indices (i.e. CCI and ECI) to be used for health assessment in the clinic and risk adjustment for outcome-based research specific to CP, notably for mortality-based research. The WCI, whether weighted or unweighted, provides improved model fit and discrimination for predicting 1- and 2-year mortality among privately insured adults with CP compared to the CCI and ECI. The WCI contains 27 comorbidities, seven of which were modified for the WCI (e.g. epilepsy as its own comorbidity) and seven of which were not part of the CCI or ECI: gastrointestinal issues; osteoarthritis; intellectual disabilities; bone fragility; dysphagia; pneumonia; and neurogenic bowel or bladder. While the added comorbidities may be uncommon or even rare for the general population, they are more prevalent and possibly more clinically relevant to health, disease, function, and mortality for adults with CP.^{1–15}

Table 1: Baseline descriptive characteristics of adults with cerebral palsy ($n=3092$)

	<i>n</i> (%)
Age, mean (SD)	48.0 (19.2)
18–40y	1136 (36.7)
41–64y	1245 (40.3)
≥65y	711 (23.0)
Sex	
Female	1511 (48.9)
Male	1581 (51.1)
Ethnic group	
White	2024 (65.5)
Black	270 (8.7)
Hispanic	259 (8.4)
Asian	86 (2.8)
Unknown/missing	453 (14.7)
US region of residence	
West	858 (27.8)
Midwest	706 (22.8)
South	1140 (36.9)
Northeast	388 (12.6)

Table 2: Prevalence of individual comorbidities from the Whitney Comorbidity Index (WCI) and association with 2-year mortality ($n=73$) among adults with cerebral palsy ($n=3092$)

	<i>n</i> (%)	Unadjusted HR (95% CI)	<i>p</i> (unadjusted competing risk)	Adjusted ^a HR (95% CI)	Adjusted ^a competing risk HR (95% CI)
<i>Included in the final WCI</i>					
Hypertension (un)complicated	1251 (40.5)	3.93 (2.35–6.58)	<0.001	1.73 (0.98–3.04)	1.74 (1.02–2.96)
Epilepsy	764 (24.7)	1.41 (0.86–2.31)	0.172	2.26 (1.37–3.73)	2.33 (1.42–3.82)
Chronic pulmonary disease	601 (19.4)	1.62 (0.97–2.70)	0.080	1.28 (0.76–2.15)	1.25 (0.73–2.13)
Depression	555 (18.0)	2.21 (1.35–3.63)	0.002	1.80 (1.09–2.96)	1.68 (1.02–2.78)
Blood loss and deficiency anemias	521 (16.9)	2.94 (1.83–4.73)	<0.001	1.86 (1.14–3.05)	1.85 (1.10–3.10)
Gastrointestinal issues	517 (16.7)	1.77 (1.05–2.99)	0.031	1.79 (1.06–3.03)	1.75 (1.03–2.97)
Osteoarthritis and allied disorders	494 (16.0)	1.49 (0.85–2.59)	0.160	0.76 (0.43–1.35)	0.76 (0.43–1.37)
Intellectual disabilities	489 (15.8)	1.26 (0.70–2.25)	0.424	1.72 (0.95–3.11)	1.76 (0.97–3.19)
Fluid and electrolyte disorders	465 (15.0)	3.94 (2.47–6.30)	<0.001	2.97 (1.84–4.79)	2.79 (1.68–4.63)
Bone fragility	454 (14.7)	2.90 (1.78–4.72)	<0.001	1.90 (1.14–3.17)	1.91 (1.14–3.19)
Cardiac arrhythmias	450 (14.6)	4.32 (2.71–6.89)	<0.001	3.00 (1.86–4.85)	2.91 (1.76–4.79)
Hypothyroidism	424 (13.7)	1.80 (1.03–3.13)	0.038	1.23 (0.69–2.17)	1.19 (0.65–2.16)
Dysphagia	408 (13.2)	4.14 (2.57–6.66)	<0.001	3.15 (1.94–5.11)	2.94 (1.76–4.91)
Cerebrovascular disease	341 (11.0)	2.00 (1.12–3.59)	0.025	1.18 (0.65–2.14)	1.15 (0.62–2.10)
Pneumonia	310 (10.0)	3.80 (2.29–6.31)	<0.001	2.84 (1.70–4.76)	2.73 (1.61–4.63)
Diabetes without chronic complications	292 (9.4)	2.49 (1.41–4.40)	0.001	1.66 (0.93–2.95)	1.64 (0.90–2.97)
Other neurological disorders	277 (9.0)	2.55 (1.43–4.57)	0.002	1.89 (1.05–3.41)	1.80 (0.98–3.30)
Renal disease	201 (6.5)	3.67 (2.05–6.57)	<0.001	1.91 (1.04–3.49)	1.80 (0.98–3.32)
Any malignancy, including lymphoma and leukemia, except malignant neoplasm of the skin	200 (6.5)	2.94 (1.59–5.47)	<0.001	1.63 (0.86–3.06)	1.58 (0.83–3.00)
Congestive heart failure	184 (6.0)	4.55 (2.58–8.03)	<0.001	2.33 (1.29–4.21)	2.12 (1.18–3.80)
Diabetes with chronic complications	156 (5.1)	5.05 (2.86–8.91)	<0.001	3.11 (1.75–5.53)	3.08 (1.69–5.61)
Neurogenic bowel or bladder	153 (5.0)	1.71 (0.74–3.93)	0.194	1.48 (0.64–3.45)	1.50 (0.62–3.64)
Mild-to-severe liver disease	122 (4.0)	2.23 (0.97–5.13)	0.056	1.90 (0.82–4.39)	1.83 (0.80–4.21)
Dementia	102 (3.3)	4.02 (1.93–8.38)	<0.001	2.01 (0.93–4.34)	1.70 (0.75–3.89)
Myocardial infarction	76 (2.5)	4.03 (1.75–9.28)	0.001	2.00 (0.85–4.68)	1.71 (0.71–4.13)
Rheumatoid arthritis and other inflammatory polyarthropathies	49 (1.6)	4.14 (1.51–11.34)	0.007	2.62 (0.95–7.21)	2.49 (0.86–7.25)
Metastatic cancer	18 (0.6)	13.4 (4.89–36.72)	<0.001	^b	^b
<i>Excluded from the final WCI</i>					
Psychoses	498 (16.1)	1.40 (0.79–2.47)	0.299	1.22 (0.69–2.15)	1.14 (0.64–2.03)
Anxiety	496 (16.0)	1.16 (0.64–2.11)	0.687	1.14 (0.62–2.08)	1.08 (0.59–1.99)
Sleep disorders	399 (12.9)	1.09 (0.56–2.12)	0.835	1.02 (0.52–1.99)	1.03 (0.52–2.01)
Autism spectrum disorder	67 (2.2)	^b	^b	^b	^b
Alcohol or drug abuse	63 (2.0)	1.48 (0.36–6.03)	0.671	2.02 (0.49–8.32)	1.80 (0.43–7.48)

^aAdjusted for age, sex, and US region of residence. ^bSample size or number of mortality cases insufficient for analysis. HR, hazard ratio; CI, confidence interval.

There were several comorbidities that were prevalent in this sample of adults with CP that were not individually associated with mortality, including psychoses (16.1%), anxiety (16.0%), and sleep disorders (12.9%). Furthermore, the presence of autism spectrum disorder was not associated with mortality, which may be due to the lower than expected prevalence of this co-occurring neurodevelopmental condition. While autism spectrum disorder did not meet the statistical criteria to be included in the WCI for this privately insured sample of adults with CP, future studies are needed to determine if this neurodevelopmental condition would add predictive value to the WCI in adults with more severe forms of CP. Although, it is important to note that the concept of a health-related index may be more complicated and nuanced than what the presence/absence of morbidities can capture for adults with CP, given their unique medical, functional, and psychosocial profiles.

A limitation of the present study is the lack of clinical CP phenotype information, such as severity of CP. Individuals with more severe forms of CP have a higher disease

and mortality burden than individuals with milder forms of CP.^{5,10} However, since severity of CP is associated with multi-morbidity,^{5,10} the WCI likely captured excess disease risk that would be tied to severity of CP and ambulatory ability and may not have significantly contributed to the prognostic value of the index measure for mortality. Future studies are needed to determine if incorporating severity of CP or other relevant CP characteristics (e.g. type of CP) could improve the already very strong WCI models for predicting 1- and 2-year mortality or other adverse outcomes. An additional limitation of this study is that the privately insured sample may reflect a healthier segment of the adult population with CP,^{1,26} thus potentially diluting the unadjusted and adjusted association between each comorbidity initially considered in the WCI and mortality. Furthermore, given the limited number of mortality cases, important comorbidities that indeed associate with health, function, disease, or survival for adults with CP may have been excluded. Future studies are needed to validate the WCI and reconsider the excluded comorbidities (e.g.

Table 3: Whitney Comorbidity Index comorbidities and modifications from the CCI and ECI

Comorbidity	Modifications
<i>From the CCI</i>	
Any malignancy, including lymphoma and leukemia, except malignant neoplasm of the skin	Added V10 codes from the ECI comorbidity 'solid tumor without metastasis'
Blood loss and deficiency anemias	Combined blood loss anemia and deficiency anemias
Cerebrovascular disease	None
Chronic pulmonary disease	Added chronic respiratory failure
Dementia	Added Alzheimer disease
Diabetes with chronic complications	None
Diabetes without chronic complications	None
Mild-to-severe liver disease	Combined mild and moderate-to-severe liver disease
Myocardial infarction	None
Renal disease	None
<i>From the CCI and ECI</i>	
Metastatic cancer	Same code from the CCI and ECI; no modifications
<i>From the ECI</i>	
Cardiac arrhythmias	None
Congestive heart failure	None
Depression	None
Epilepsy	This was removed from the ECI comorbidity 'other neurological disorders'
Fluid and electrolyte disorders	None
Hypertension (un)complicated	None
Hypothyroidism	None
Other neurological disorders	Removed epilepsy
Rheumatoid arthritis and other inflammatory polyarthropathies	Kept the rheumatoid arthritis and other inflammatory polyarthropathies from the ECI comorbidity 'rheumatoid arthritis/collagen vascular diseases'
<i>Added (not from the CCI or ECI)</i>	
Bone fragility	
Dysphagia	
Gastrointestinal issues	
Intellectual disabilities	
Neurogenic bowel or bladder disorders	
Osteoarthritis and allied disorders	
Pneumonia	

CCI, Charlson Comorbidity Index; ECI, Elixhauser Comorbidity Index.

anxiety, sleep disorders) in a larger sample of adults with CP, such as the Medicare or Medicaid population. Lastly, claims data can contain errors in coding, such as accidental miscoding of conditions.

In conclusion, the WCI, which includes clinically relevant comorbidities for adults with CP, provides a better model fit and discrimination for predicting 1- and 2-year mortality among privately insured adults with CP compared to the CCI and ECI. Future work is needed to validate the WCI in different segments of the population with CP. We encourage researchers to use or test the WCI for disease risk adjustment in their own CP-related research.

Table 4: Model fit and discrimination statistics after further adjustment of the base model for each index separately to predict 1- and 2-year mortality among adults with cerebral palsy ($n=3092$)

	1-year mortality		2-year mortality	
	AIC	Uno's C-statistic (95% CI)	AIC	Uno's C-statistic (95% CI)
Base model ^a	780	0.78 (0.71–0.85)	1098	0.75 (0.70–0.80)
<i>As continuous</i>				
Charlson Comorbidity Index	757	0.80 (0.71–0.89)	1074	0.77 (0.72–0.83)
Elixhauser Comorbidity Index	761	0.81 (0.75–0.87)	1068	0.75 (0.68–0.82)
Whitney Comorbidity Index				
Unweighted	750	0.83 (0.77–0.89)	1058	0.81 (0.76–0.85)
Weighted	739	0.84 (0.78–0.90)	1047	0.81 (0.76–0.87)
<i>As discrete</i>				
Charlson Comorbidity Index	770	0.81 (0.74–0.88)	1082	0.79 (0.72–0.85)
Elixhauser Comorbidity Index	774	0.82 (0.73–0.91)	1080	0.80 (0.74–0.87)
Whitney Comorbidity Index				
Unweighted	754	0.85 (0.78–0.92)	1061	0.83 (0.76–0.90)
Weighted	748	0.88 (0.82–0.94)	1065	0.84 (0.76–0.91)

^aAdjusted for age, sex, and US region of residence. AIC, Akaike Information Criterion; CI, confidence interval.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from Optum Clinformatics® Data Mart Database. Restrictions apply to the availability of these data, which were used under license for this study.

SUPPORTING INFORMATION

The following additional material may be found online:

Table S1: ICD-9-CM codes for each comorbidity initially considered for the WCI

Table S2: Prevalence of individual comorbidities from the CCI and association with 2-year mortality among adults with CP

Table S3: Prevalence of individual comorbidities from the ECI and association with 2-year mortality among adults with CP

Table S4: Model fit and discrimination statistics after further adjusting the base model for each index separately for predicting 2-year mortality among adults with CP with complete data on ethnic grouping

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