Validation of the Early Onset Scoliosis Questionnaire (EOSQ) as applied to the Classification of Early Onset Scoliosis (C-EOS) Etiology Designation Before Scoliosis Treatment

Brandon Ramo, Anna McClung, Chan-Hee Jo , Burt Yazsay, Lindsay Andras, Matt Oetgen, Pediatric Spine Study Group

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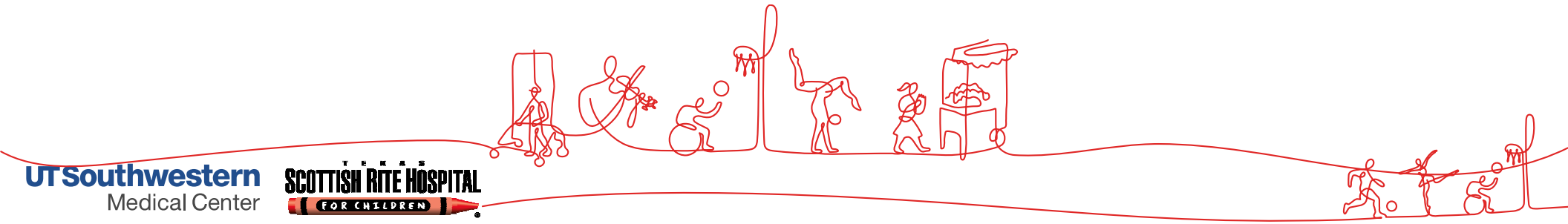
PEDIATRIC SPINE STUDY GROUP



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Disclosures

- Brandon Ramo: Nuvasive, Inc; Orthopediatrics: Speaker's Bureau
- Anna McClung: none
- Chan-Hee Jo: none
- Burt Yazsay: Depuy Synthes - consultant and speakers bureau; Stryker/K2M - consultant, speakers bureau, royalties; Nuvasive - consultant, speakers bureau, royalties; Globus – royalties; OrthoPediatrics – royalties; Biogen - consultant
- Lindsay Andras: Biomet: Speaker's Bureau, Eli Lilly: Stock/Shareholder; POSNA, JPO, Scoliosis Research Society: Editorial/Governing Board Nuvasive, Medtronic: Speakers bureau; Orthobullets: Other support
- Matt Oetgen: none
- PSSG: Research support: POSNA, FDA, NuVasive, DePuy Synthes Spine, Growing Spine Foundation, Children's Spine Foundation

Background

- First Disease Specific Patient (Parent)-Reported Outcomes Questionnaire for EOS

- Validity
  - Criterion Validity (pulm) (n=10)
  - Construct Validity (n=95)
- Reliability
  - n=15
- Responsiveness
  - n=25 pre and post-operative
- Normative Reference Data (cross-sectional)
  - 150 norms (benign orthopaedic dx's)

ORIGINAL ARTICLE

Measuring Quality of Life in Children With Early Onset Scoliosis: Development and Initial Validation of the Early Onset Scoliosis Questionnaire

Jacqueline Corona, MD,† Hiroko Matsumoto, MA,*† David P. Roye, Jr, MD,*† and Michael G. Vitale, MD, MPH*†*

(*J Pediatr Orthop* 2011;31:180–185)

The Final 24-Item Early Onset Scoliosis Questionnaires (EOSQ-24): Validity, Reliability and Responsiveness

Hiroko Matsumoto, MA,† Brendan Williams, MD,‡ Howard Y. Park, MD,§ Julie Y. Yoshimachi, BA,* Benjamin D. Roye, MD, MPH,* David P. Roye, Jr, MD,* Behrooz A. Akbarnia, MD,‖ John Emans, MD,¶ David Skaggs, MD,§# John T. Smith, MD,** and Michael G. Vitale, MD, MPH**

(*J Pediatr Orthop* 2018;38:144–151)

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Background

- HRQOL 8 domains:
  - General Health
  - Pain/Discomfort
  - Pulmonary Function
  - Transfer
  - Physical Function
  - Daily Living
  - Fatigue/Energy Level
  - Emotion
- Family Burden 2 domains:
  - Parental Impact
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Known-Group Validity

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TABLE 2. Known Group Validity: The EOSQ-24 Mean Scores Based on Etiology

	Mean (95% CI)				
	Idiopathic (N = 29)	**Congenital/Structural (N = 15)**	**Syndromic (N = 18)**	**Neuromuscular (N = 28)**	***P***
General health	80.6[a] (74.3-86.9)	80.8[b] (73.0-88.6)	62.5[c] (51.2-73.8)	64.4[d] (56.2-72.5)	a vs. b = 0.999 **a vs. c = 0.010** **a vs. d = 0.010** **b vs. c = 0.034** **b vs. d = 0.040** c vs. d = 0.988
Pain/fatigue	79.3[a] (70.2-88.4)	76.7[b] (62.6-90.7)	72.8[c] (61.6-84.0)	70.8[d] (63.0-78.7)	a vs. b = .983 a vs. c = 0.781 a vs. d = 0.502 b vs. c = 0.963 b vs. d = 0.854 c vs. d = 0.992
Pulmonary function	92.9[a] (87.5-98.2)	92.5[b] (86.2-98.8)	77.3[c] (64.0-90.7)	78.8[d] (67.7-89.9)	a vs. b = 0.999 a vs. c = 0.070 a vs. d = 0.066 b vs. c = 0.156 b vs. d = 0.171 c vs. d = 0.996
Transfer	89.3[a] (79.6-99.0)	96.4[b] (88.7-104.1)	85.3[c] (74.1-96.5)	63.5[d] (52.3-74.6)	a vs. b = 0.795 a vs. c = 0.947 **a vs. d = < 0.001** b vs. c = 0.567 **b vs. d = < 0.001** **c vs. d = 0.021**

Total N = 90 patients

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Background

- C-EOS

Development and Initial Validation of the Classification of Early-Onset Scoliosis (C-EOS)

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Investigation performed at Columbia University Medical Center, New York, NY

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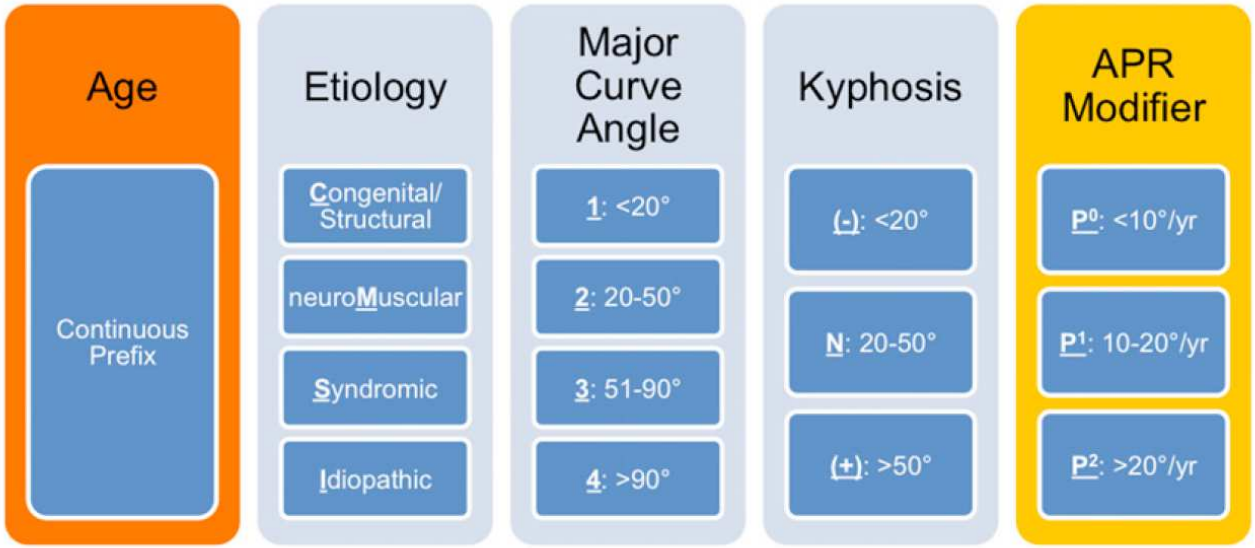
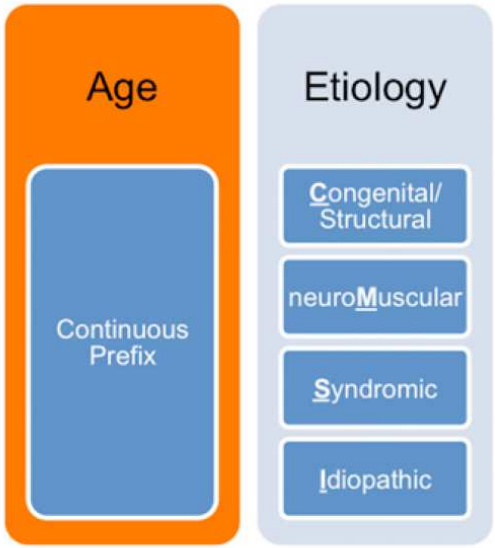


Background

- C-EOS



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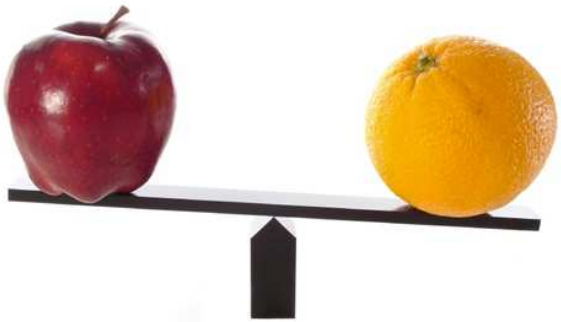

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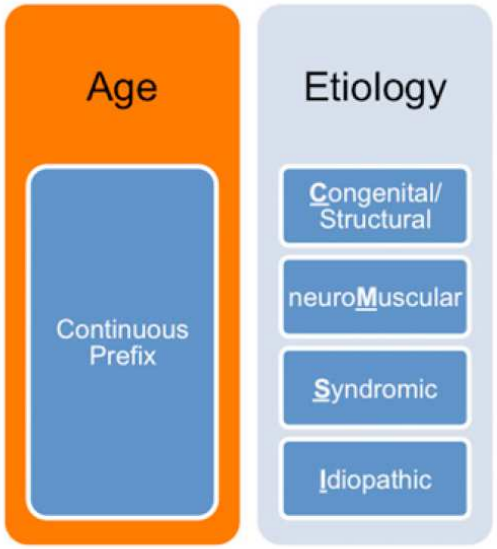

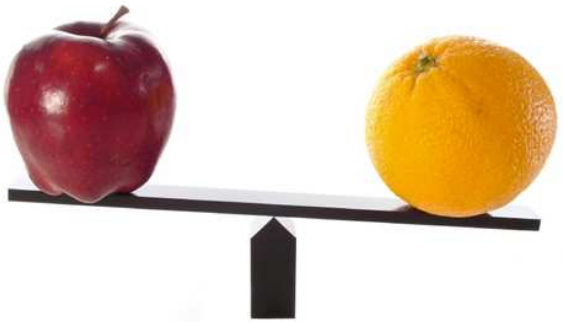

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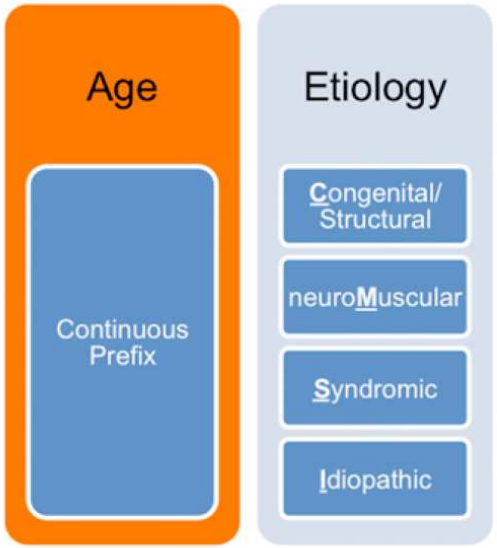

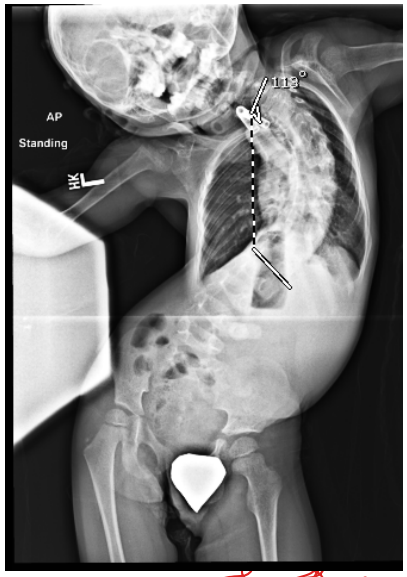

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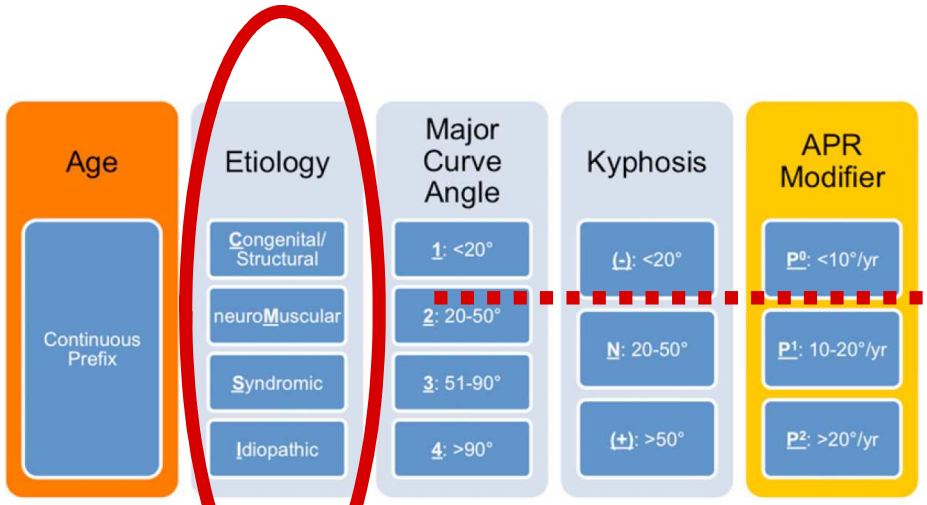
Hypotheses:

- If the C-EOS is a valid way to distinguish patients from one another, then there should be differences between etiology designations in EOSQ domain scores prior to intervention in EOS patients



- HRQOL 8 domains:
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  - Transfer
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  - Fatigue/Energy Level
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  - Parental Impact
  - Financial Impact
- Satisfaction:
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Methods

- Retrospective comparative analysis of prospectively collected data of a large multi-center cohort of patients.
- 610 patients who had pre-treatment (surgical or non-operative) EOSQ
  - 119 congenital
  - 201 idiopathic
  - 156 neuromuscular
  - 184 syndromic

Baseline Characteristics

C-EOS Etiology					
	All n=610	Idiopathic n=201	Congenital n=119	Syndromic n=134	Neuromuscular n=156
Age at Baseline/Pre-Treatment EOSQ	6.1 ± 3.8	6.3 ± 4.7	4.5 ± 3.2	6.2 ± 3.4	7.0 ± 2.7
Coronal Cobb Angle	63 ± 25	54 ± 22	56 ± 23	67 ± 23	76 ± 26
Max Sagittal Kyphosis	48 ± 25	41 ± 22	41 ± 20	52 ± 26	57 ± 27

Neuromuscular patients were slightly older.
Neuromuscular and Syndromic Patients had slightly larger coronal and sagittal Cobbs

Results

Table 1. Summary Statistics for All and by DX Group

	All		C-EOS DX									Multiple Comparisons					
			Cong (1)		IS (2)		NM (3)		Synd (4)								
Variable	n	mean ± std, med (range)	n	mean ± std, med (range)	n	mean ± std, med (range)	n	mean ± std, med (range)	n	mean ± std, med (range)	pValue	1 vs. 2	1 vs. 3	1 vs. 4	2 vs. 3	2 vs. 4	3 vs. 4
Ageattreatment	610	6.12 ± 3.76,5.78 (-0.64, 16.76)	119	4.49 ± 3.16,4.16 (-0.22, 14.7)	201	6.32 ± 4.67,5.02 (0.59, 16.76)	156	7.03 ± 2.66,7.09 (1.07, 12.87)	134	6.23 ± 3.36,5.86 (-0.64, 13.8)	0.0000	0.0033	0.0000	0.0000	0.0147	0.4742	0.0191
PrimaryCurveDegree	530	62.9 ± 24.75,60.5 (4, 143)	93	56.3 ± 23.2,52 (12, 126)	183	54.27 ± 21.55,52 (17, 143)	138	75.51 ± 25.61,79.5 (4, 130)	116	66.79 ± 22.6,69.5 (7, 141)	0.0000	0.5940	0.0000	0.0003	0.0000	0.0000	0.0020
Abs_GK	360	48.56 ± 24.97,45 (0, 135)	78	41.35 ± 19.83,39.74 (8, 106)	87	41.02 ± 21.68,38 (5, 110)	104	57.03 ± 27.3,53.5 (0, 135)	91	52.27 ± 25.51,48 (1, 114)	0.0000	0.7839	0.0001	0.0041	0.0000	0.0024	0.2576
majorCurve	534	65.88 ± 24.97,64 (14, 143)	96	58.56 ± 23.16,53.83 (14, 126)	183	55.35 ± 21.58,52 (17, 143)	138	80.87 ± 24.01,84.5 (24, 135)	117	70.69 ± 22.43,72 (23, 141)	0.0000	0.3114	0.0000	0.0001	0.0000	0.0000	0.0004
GeneralHealth	610	71.7 ± 19.4,75 (0, 100)	119	75 ± 19.53,75 (12.5, 100)	201	77.24 ± 16.67,75 (25, 100)	156	65.38 ± 19.92,62.5 (0, 100)	134	67.82 ± 19.65,75 (0, 100)	0.0000	0.4550	0.0001	0.0022	0.0000	0.0000	0.3683
PainDiscomfort	601	74.63 ± 22.65,75 (12.5, 100)	118	81.04 ± 20.85,75 (25, 100)	201	76.68 ± 21.85,75 (12.5, 100)	151	67.38 ± 24.62,62.5 (12.5, 100)	131	74.05 ± 20.89,75 (12.5, 100)	0.0000	0.0782	0.0000	0.0046	0.0003	0.1741	0.0258
PulmonaryFunction	604	85.37 ± 22.65,100 (0, 100)	117	87.93 ± 20.56,100 (0, 100)	201	91.79 ± 15.19,100 (25, 100)	154	78.57 ± 27.81,100 (0, 100)	132	81.25 ± 24.3,87.5 (0, 100)	0.0000	0.2845	0.0061	0.0098	0.0000	0.0000	0.8027
Transfer	601	78.08 ± 28.19,100 (0, 100)	118	86.23 ± 22.79,100 (0, 100)	200	91.38 ± 17.29,100 (25, 100)	152	59.7 ± 31.39,50 (0, 100)	131	71.76 ± 28.82,75 (0, 100)	0.0000	0.0573	0.0000	0.0000	0.0000	0.0000	0.0011
PhysicalFunction	595	69.24 ± 33.65,83.33 (0, 100)	115	82.93 ± 25.15,100 (0, 100)	201	84.02 ± 23.75,100 (0, 100)	149	40.94 ± 35.14,33.33 (0, 100)	130	66.73 ± 30.41,75 (0, 100)	0.0000	0.7543	0.0000	0.0000	0.0000	0.0000	0.0000
DailyLiving	586	59.92 ± 36.04,62.5 (0, 100)	114	73.46 ± 30.03,87.5 (0, 100)	200	79.81 ± 26.78,93.75 (0, 100)	143	30.59 ± 31.74,25 (0, 100)	129	49.61 ± 32.51,50 (0, 100)	0.0000	0.0420	0.0000	0.0000	0.0000	0.0000	0.0000
FatigueEnergy	602	73.19 ± 27.35,75 (0, 100)	117	82.05 ± 23.75,100 (12.5, 100)	200	84.94 ± 20.56,100 (25, 100)	152	56.74 ± 29.12,56.25 (0, 100)	133	66.54 ± 25.82,75 (0, 100)	0.0000	0.4975	0.0000	0.0000	0.0000	0.0000	0.0042
Emotion	584	75.68 ± 23.83,75 (0, 100)	113	82.96 ± 22.04,87.5 (0, 100)	200	78.44 ± 22.45,87.5 (0, 100)	144	68.49 ± 25.65,75 (0, 100)	127	73.03 ± 22.95,75 (0, 100)	0.0000	0.0291	0.0000	0.0001	0.0003	0.0245	0.1762
ParentalImpact	604	66.92 ± 23.96,70 (0, 100)	119	75.47 ± 22.26,80 (5, 100)	199	75.83 ± 21.12,80 (15, 100)	153	53.59 ± 23.71,50 (0, 100)	133	61.26 ± 20.97,60 (16.67, 100)	0.0000	0.9263	0.0000	0.0000	0.0000	0.0000	0.0046
FinancialImpact	595	71.26 ± 30.28,75 (0, 100)	118	76.69 ± 28.63,87.5 (0, 100)	198	76.39 ± 27.27,75 (0, 100)	148	62.67 ± 33.18,75 (0, 100)	131	68.32 ± 30.35,75 (0, 100)	0.0001	0.7680	0.0004	0.0189	0.0001	0.0157	0.1863
ChildSatisfaction	567	68.03 ± 27.64,75 (0, 100)	108	78.7 ± 24.12,75 (25, 100)	197	77.16 ± 24.19,75 (0, 100)	138	52.17 ± 28.73,50 (0, 100)	124	61.9 ± 24.58,75 (0, 100)	0.0000	0.5242	0.0000	0.0000	0.0000	0.0000	0.0073
ParentSatisfaction	578	67.91 ± 28.34,75 (0, 100)	112	77.46 ± 24.19,75 (0, 100)	198	76.52 ± 25.27,75 (0, 100)	142	55.28 ± 30.19,50 (0, 100)	126	60.12 ± 26.68,62.5 (0, 100)	0.0000	0.8273	0.0000	0.0000	0.0000	0.0000	0.1989

Please note that we construct subdomain based on the instruction of 'less than 50% missing' of the items. All the domains were converted to standardized 0-100 scales.

A P-value for 4 groups comparisons was calculated using a Kruskal-Wallis test, followed by Mann-Whitney test for two groups. A Bonferroni adjustment can be used for multiple comparisons.

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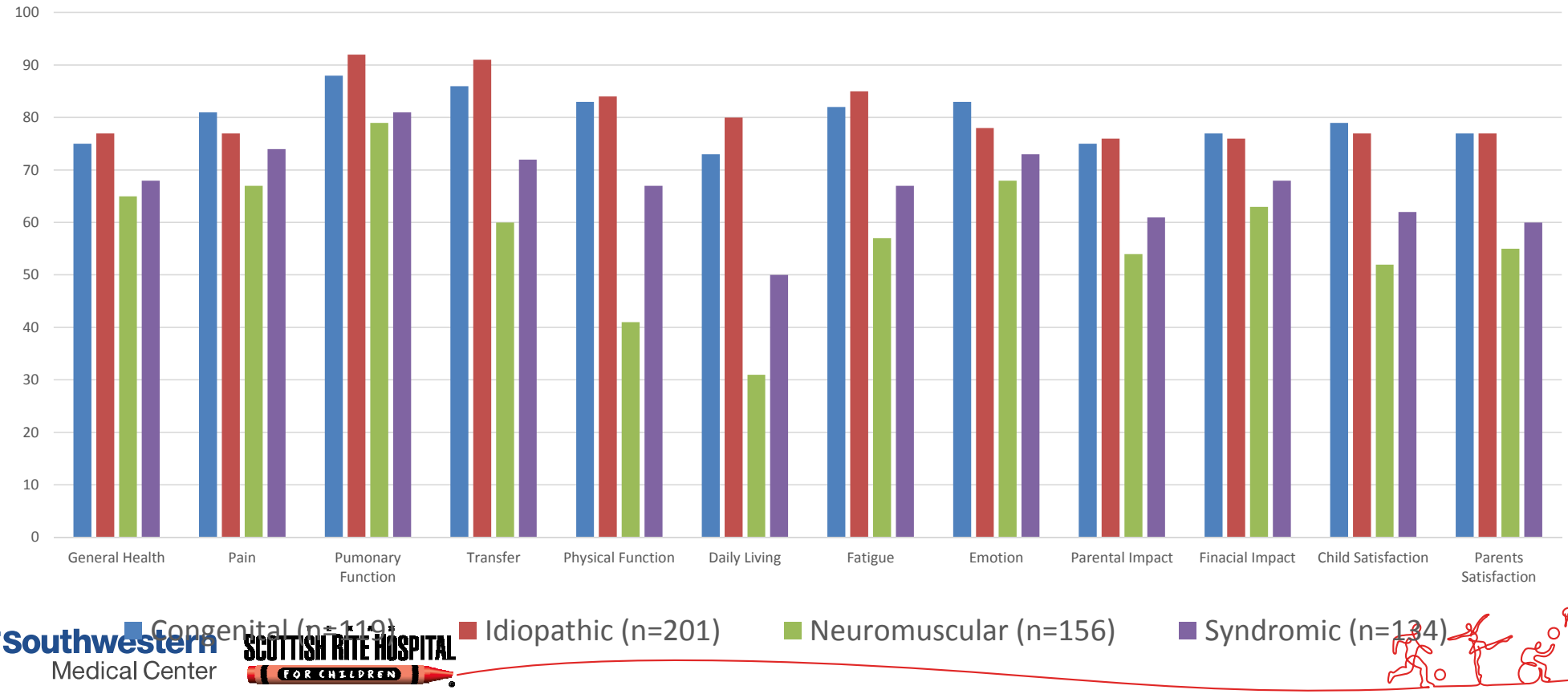
EOSQ Domains based on C-EOS Diagnosis



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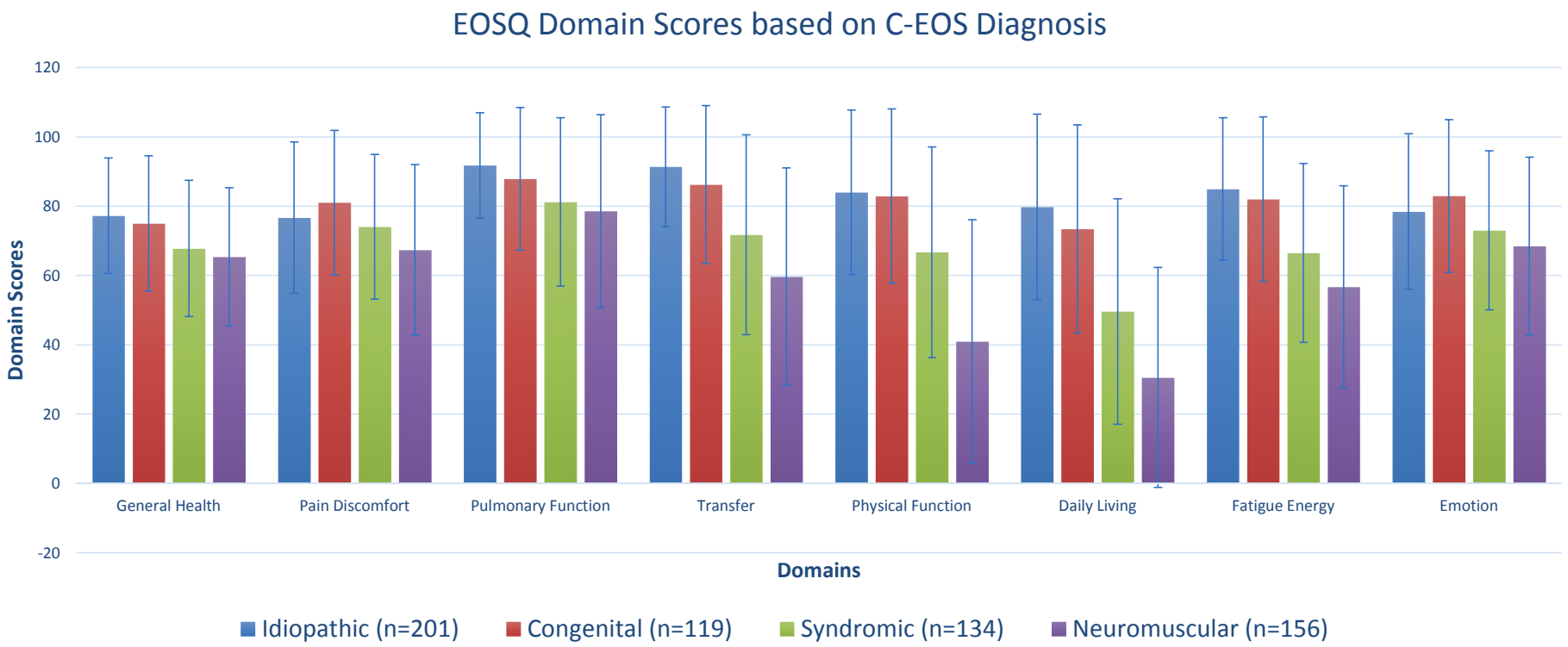
Results


EOSQ Domain Scores based on C-EOS Diagnosis
120
100
80
60
40
20
0
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Domain Scores
General Health
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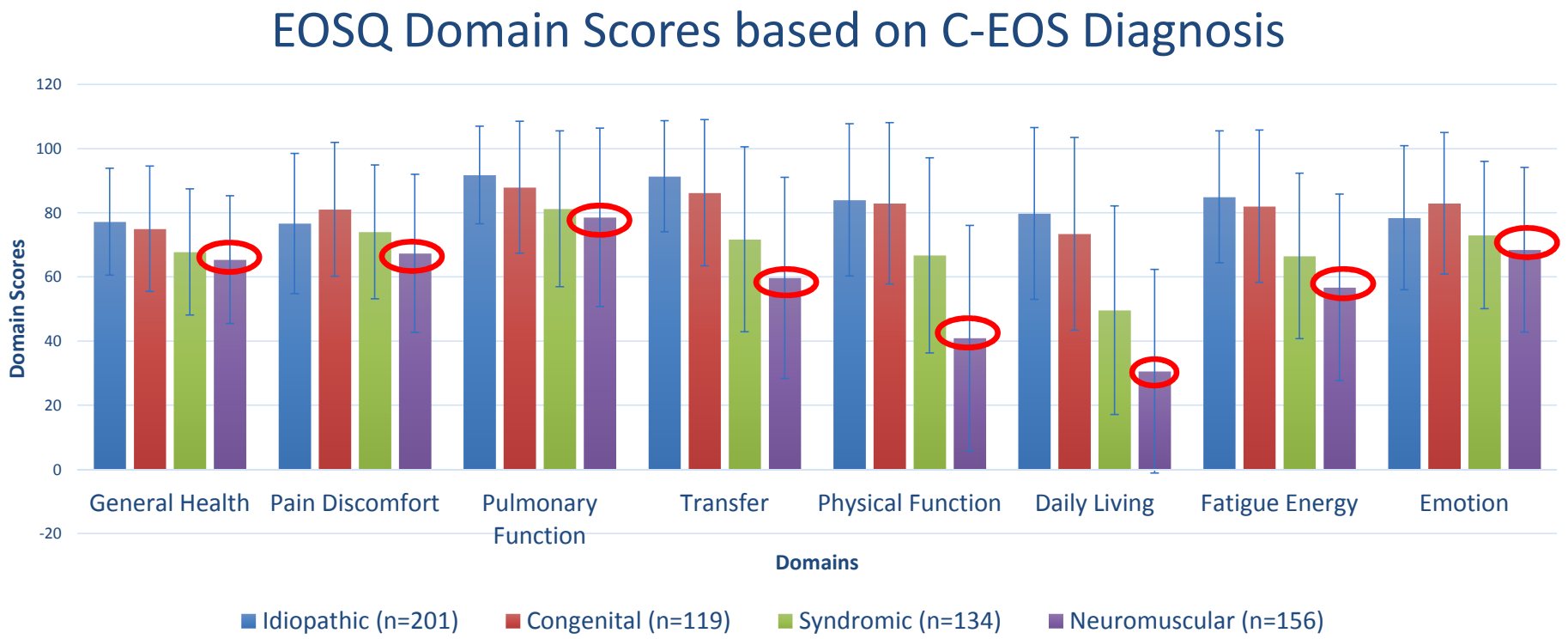
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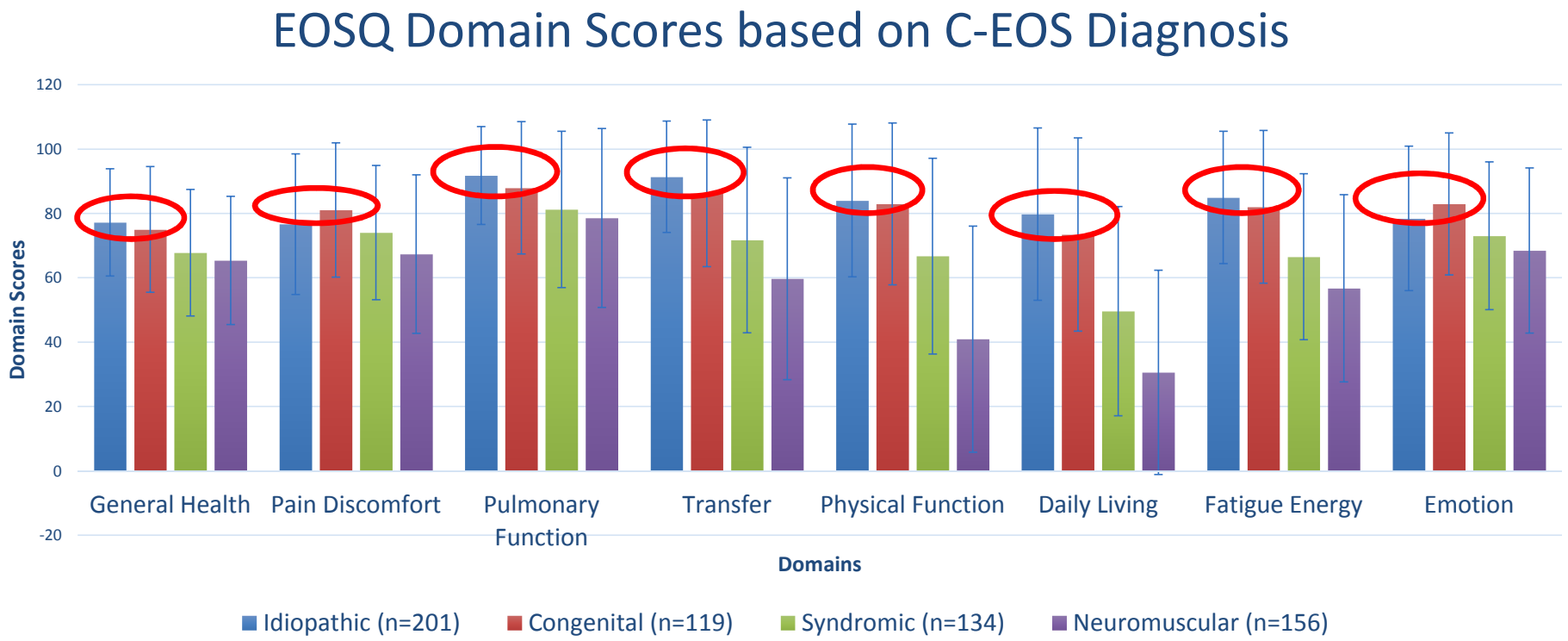

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Results – Multivariate Summary

Variables affecting EOSQ Domain Scores in Multivariate Modeling												
	General Health	Pain	Pulmonary Function	Transfer	Physical Function	Daily Living	Fatigue/ Energy	Emotion	Parental Impact	Financial impact	Parent Satisfaction	Child Satisfaction
Age		(-)			(+)	(+)		(-)				
C-EOS	C > S,NM		I>S	C, I > NM, S	C > NM	C, I > NM, S	C, I > NM, S	C,I > NM,S	C > S		C >NM	C > NM,S
Ambulatory Status		(+)		(+)	(+)	(+)	(+)		(+)			
Cardiac Condition												
Pulmonary Condition	(-)		(-)									
Tracheostomy			(-)									
Supplemental nutrition						(-)						
Developmental Delay					(-)	(-)					(-)	
GI condition						(-)						
(+) implies positive correlation with subdomain score, (-) implies negative correlation with subdomain score												

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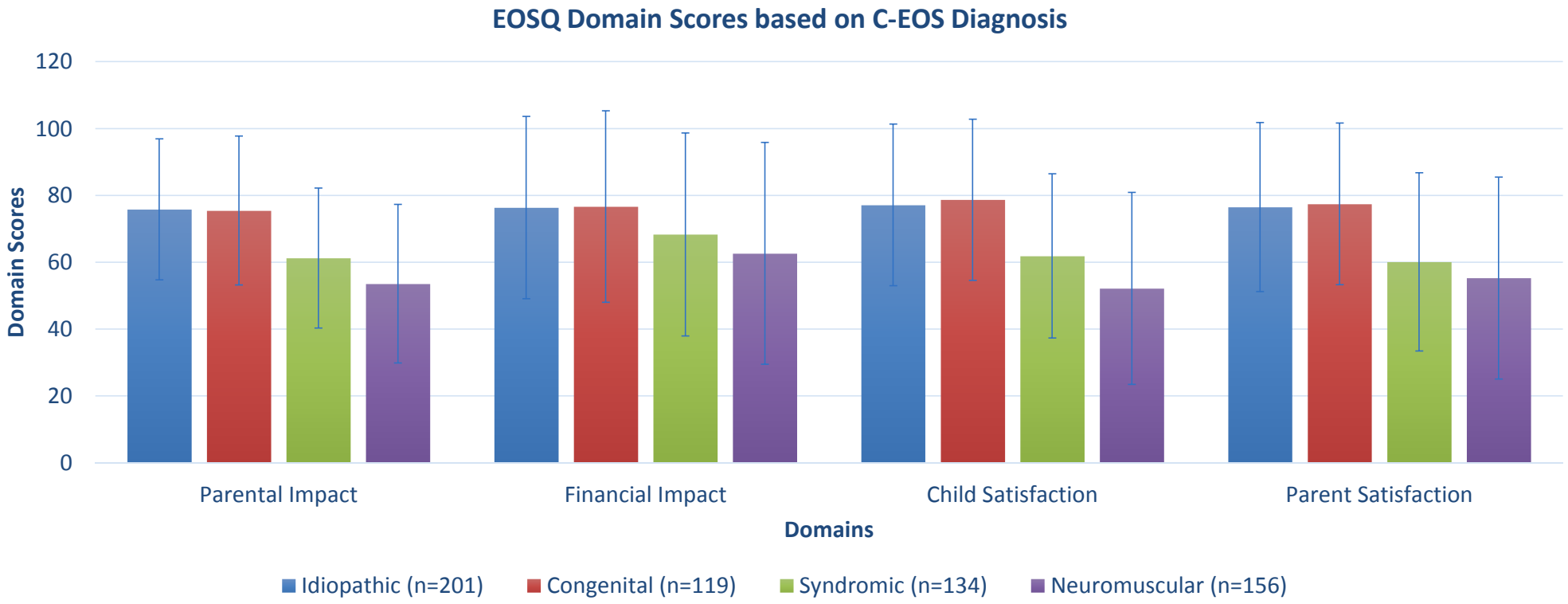

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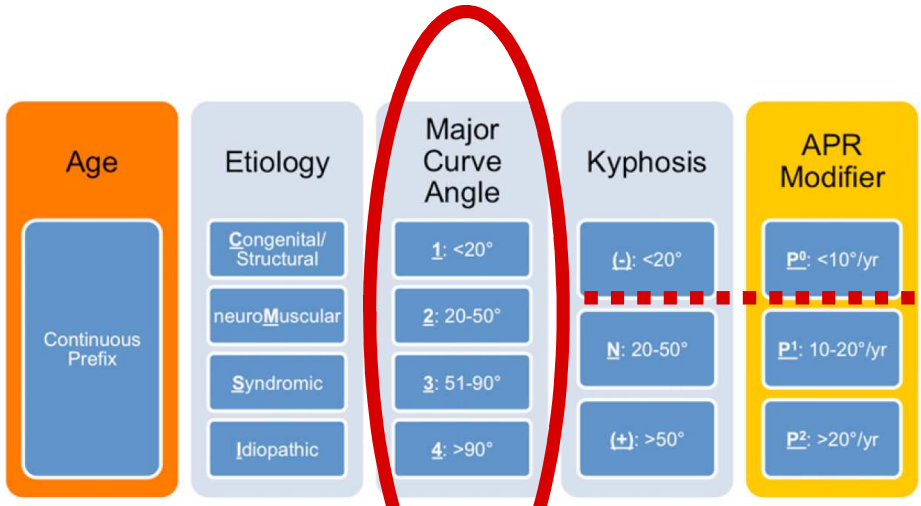
Interpretation:

- Idiopathic = Congenital
  - Effectively no domains where congenital scored differently than idiopathic.

- There was no domain where neuromuscular or syndromic patients scored as well as congenital or idiopathic
  - except pain where syndromic caught up to idiopathic

- Neuromuscular patients generally scored the lowest in EVERY domain.
  - A few exceptions where the syndromic patients were similarly poor such as general health, pulmonary function, emotion, financial impact and parent satisfaction

Hypotheses:



- HRQOL 8 domains:
  - General Health
  - Pain/Discomfort
  - Pulmonary Function
  - Transfer
  - Physical Function
  - Daily Living
  - Fatigue/Energy Level
  - Emotion
- Family Burden:
  - Parental Impact
  - Financial Impact
- Satisfaction:
  - Child Satisfaction
  - Parent Satisfaction

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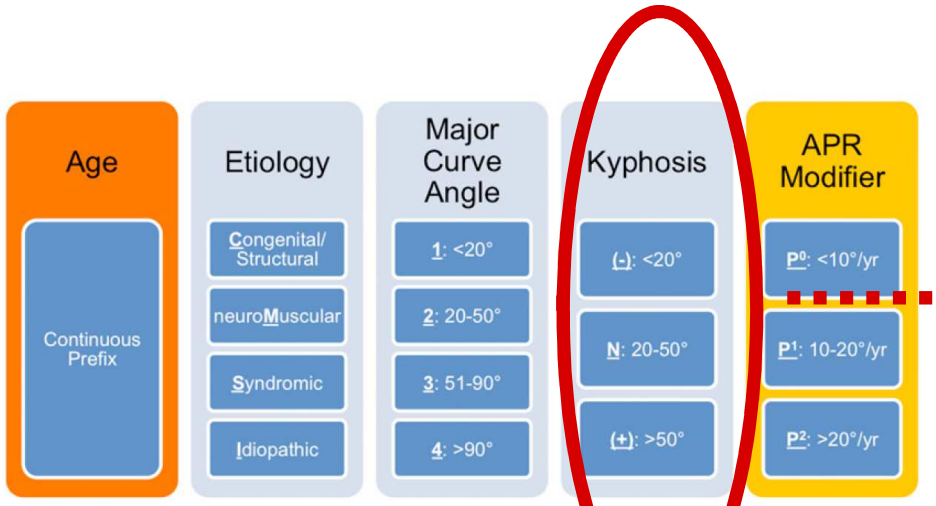
Coronal Cobb angles had a statistically significant effect on all domains with a negative correlation

BUT the effects were small with low Spearman coefficients

Coronal Cobb Angle			
	n	Spearman correlation	p-value
General Health	530	-0.109	0.012
Pain Discomfort	524	-0.233	0.000
Pulmonary Function	524	-0.177	0.000
Transfer	522	-0.288	0.000
Physical Function	517	-0.258	0.000
Daily Living	514	-0.272	0.000
Fatigue Energy	524	-0.247	0.000
Emotion	509	-0.203	0.000
Parental Impact	525	-0.272	0.000
Financial Impact	518	-0.209	0.000
Child Satisfaction	495	-0.241	0.000
Parent Satisfaction	502	-0.211	0.000

Hypotheses:



- HRQOL 8 domains:
  - General Health
  - Pain/Discomfort
  - Pulmonary Function
  - Transfer
  - Physical Function
  - Daily Living
  - Fatigue/Energy Level
  - Emotion
- Family Burden:
  - Parental Impact
  - Financial Impact
- Satisfaction:
  - Child Satisfaction
  - Parent Satisfaction

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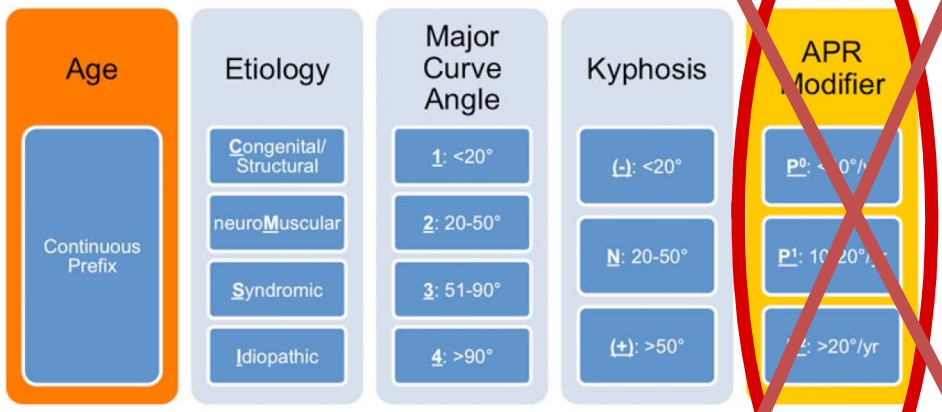
Kyphosis angles had a statistically significant effect on all domains with a negative correlation

BUT the effects were small with low Spearman coefficients

Maximum Kyphosis			
	n	Spearman correlation	p-value
General Health	360	-0.094	0.074
Pain Discomfort	355	-0.138	0.009
Pulmonary Function	354	-0.163	0.002
Transfer	352	-0.139	0.009
Physical Function	352	-0.223	0.000
Daily Living	345	-0.196	0.000
Fatigue Energy	356	-0.242	0.000
Emotion	339	-0.123	0.024
Parental Impact	356	-0.206	0.000
Financial Impact	351	-0.141	0.008
Child Satisfaction	330	-0.201	0.000
Parent Satisfaction	334	-0.202	0.000

Hypotheses:



Age

- HRQOL 8 domains:
  - General Health
  - Pain/Discomfort
  - Pulmonary Function
  - Transfer
  - Physical Function
  - Daily Living
  - Fatigue/Energy Level
  - Emotion
- Family Burden:
  - Parental Impact
  - Financial Impact
- Satisfaction:
  - Child Satisfaction
  - Parent Satisfaction

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Looked at variables which could create bias

- Age had a statistically significant effect on several domains but the actual effects were small with low Spearman coefficients

	Age		
	N	Corr	P-val
General Health	610	0.042	0.302
Pain Discomfort	601	-0.222	0.000
Pulmonary Function	604	-0.102	0.012
Transfer	601	-0.065	0.112
Physical Function	595	0.039	0.348
Daily Living	586	0.058	0.164
Fatigue Energy	602	-0.158	0.000
Emotion	584	-0.246	0.000
Parental Impact	604	-0.078	0.055
Financial Impact	595	-0.028	0.493
Child Satisfaction	567	-0.119	0.004
Parent Satisfaction	578	-0.081	0.051

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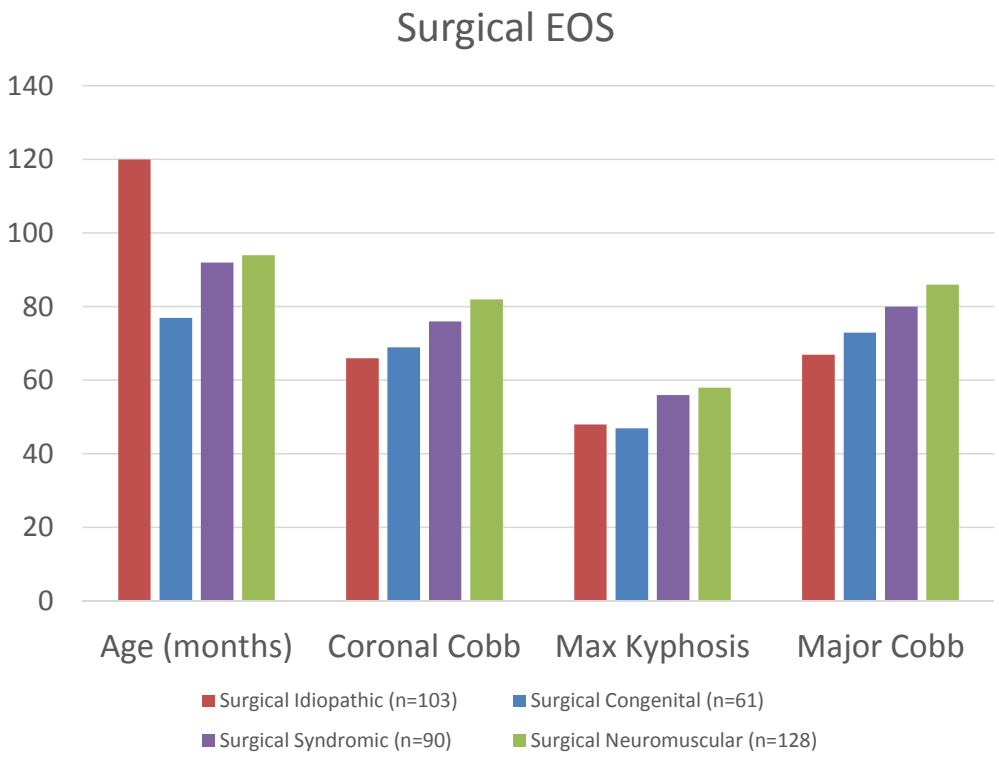
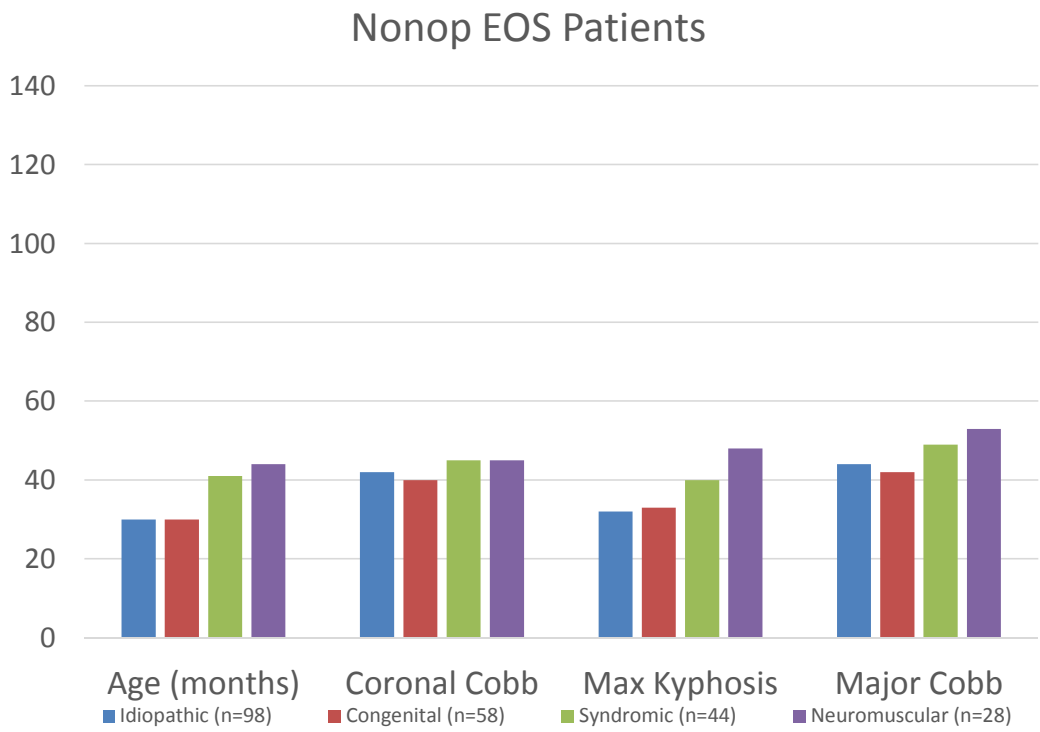
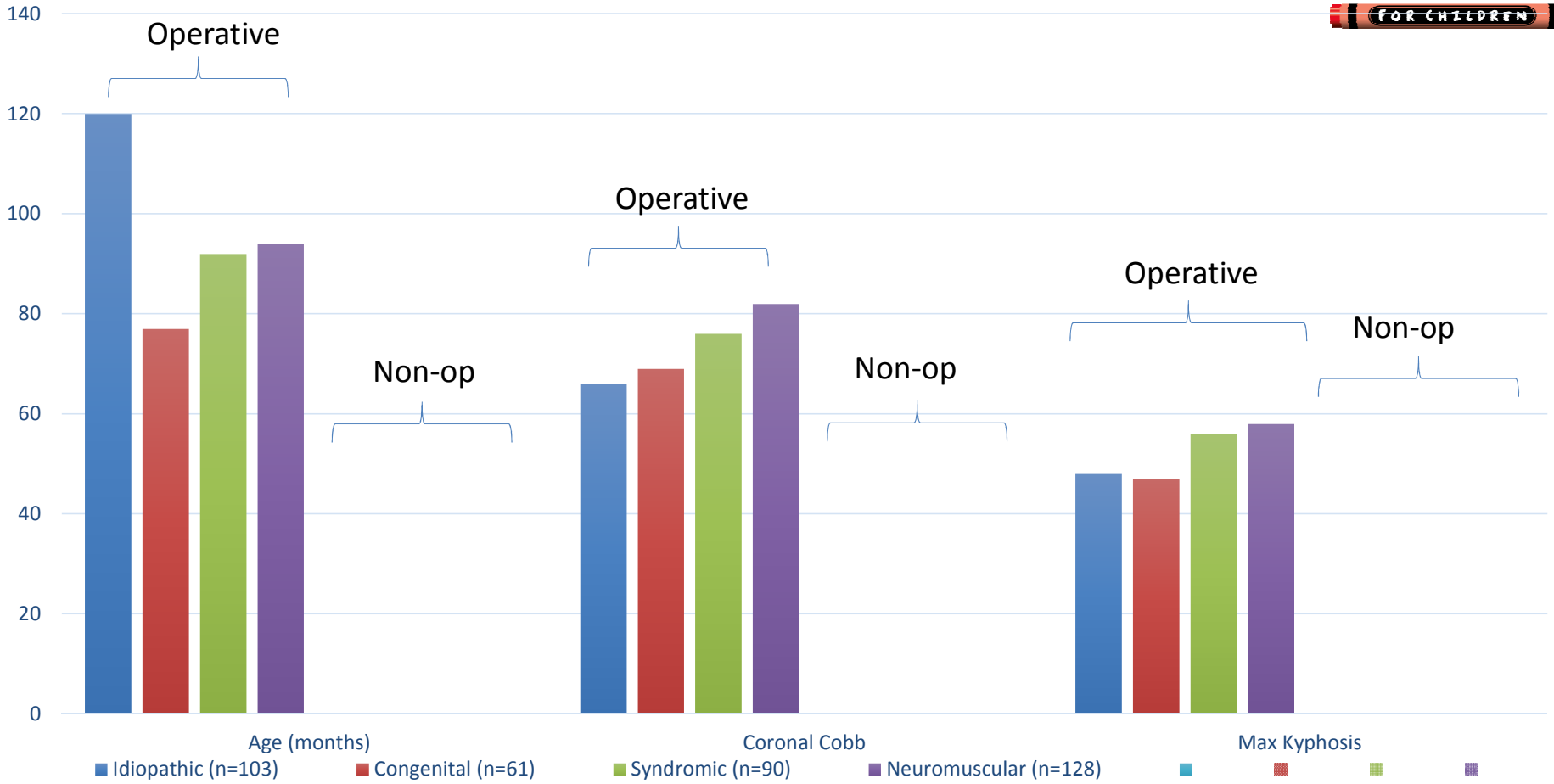
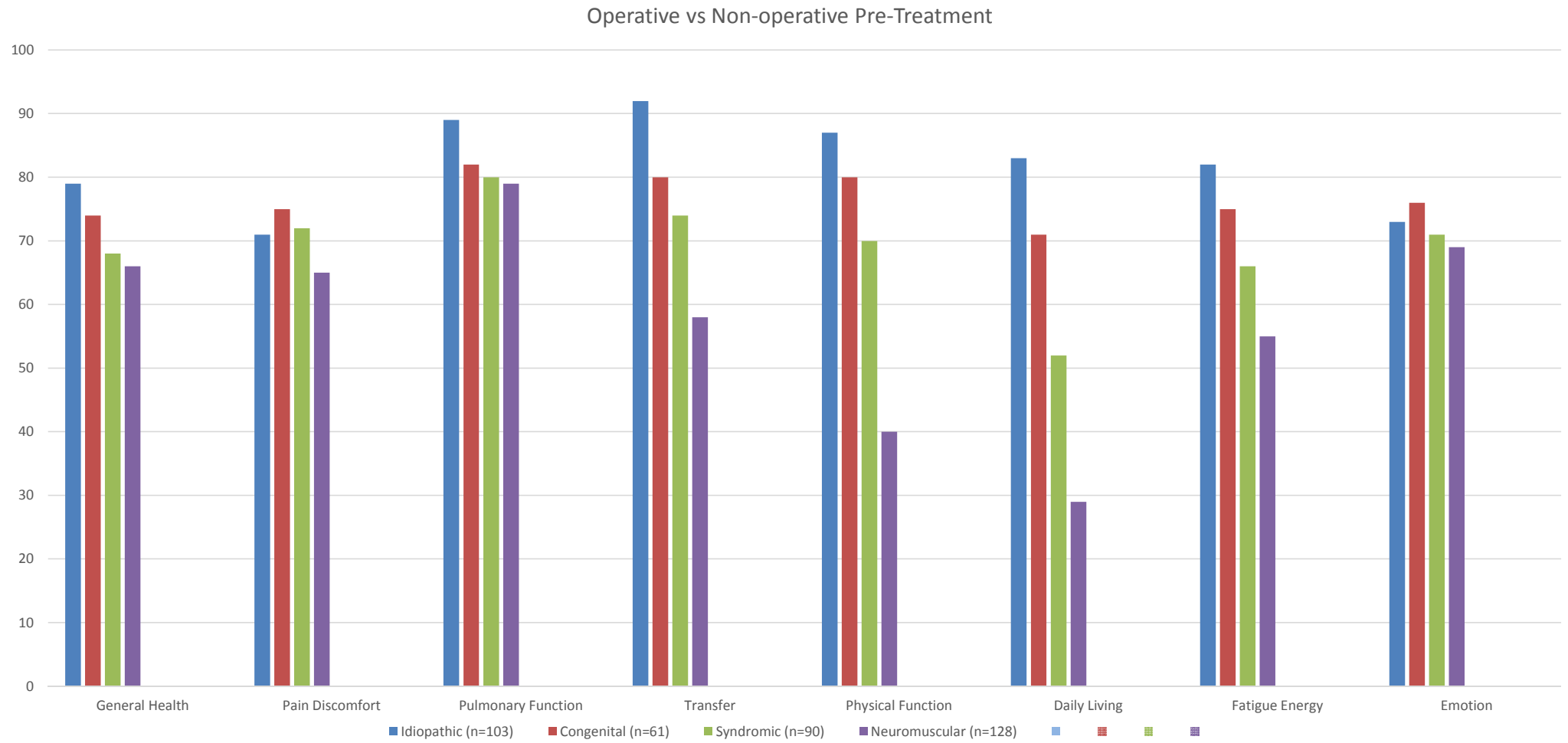

Surgical EOS
140
120
100
80
60
40
20
0
Age (months)
Coronal Cobb
Max Kyphosis
Major Cobb
Surgical Idiopathic (n=103)
Surgical Congenital (n=61)
Surgical Syndromic (n=90)
Surgical Neuromuscular (n=128)


Nonop EOS Patients
140
120
100
80
60
40
20
0
Age (months)
Coronal Cobb
Max Kyphosis
Major Cobb
Idiopathic (n=98)
Congenital (n=58)
Syndromic (n=44)
Neuromuscular (n=28)

Operative vs Non-operative Patients (all pre-treatment)





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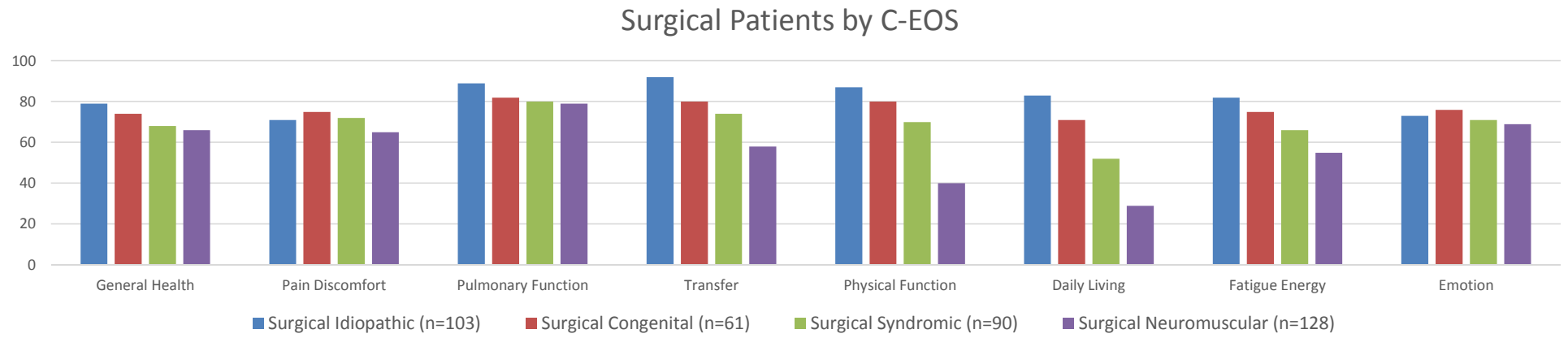
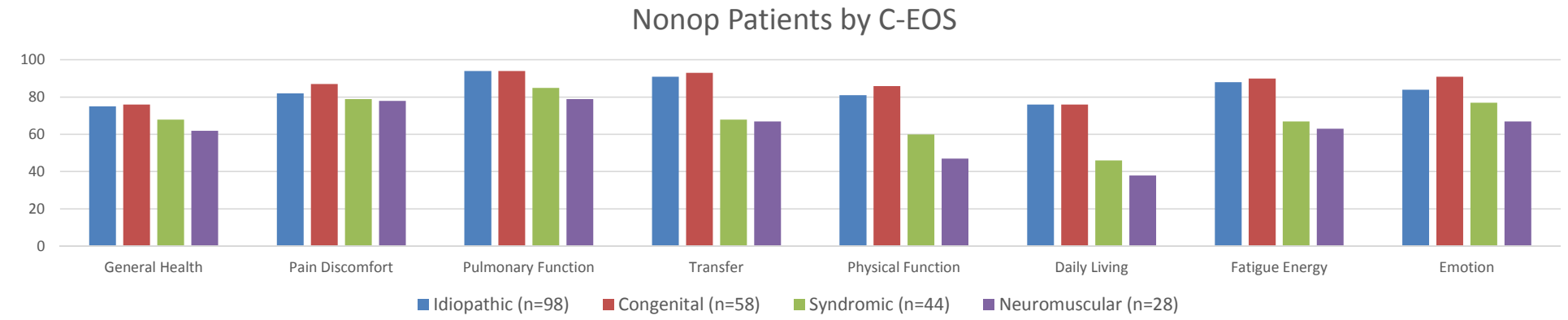
But similar effects on scores, nonop or operative:





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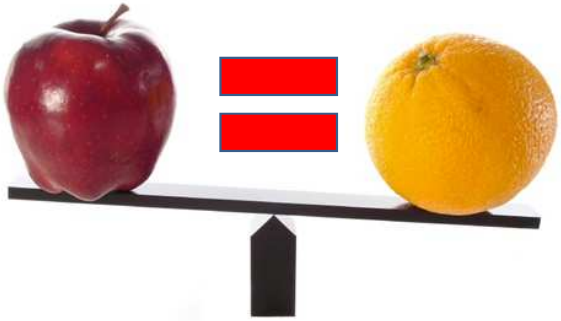
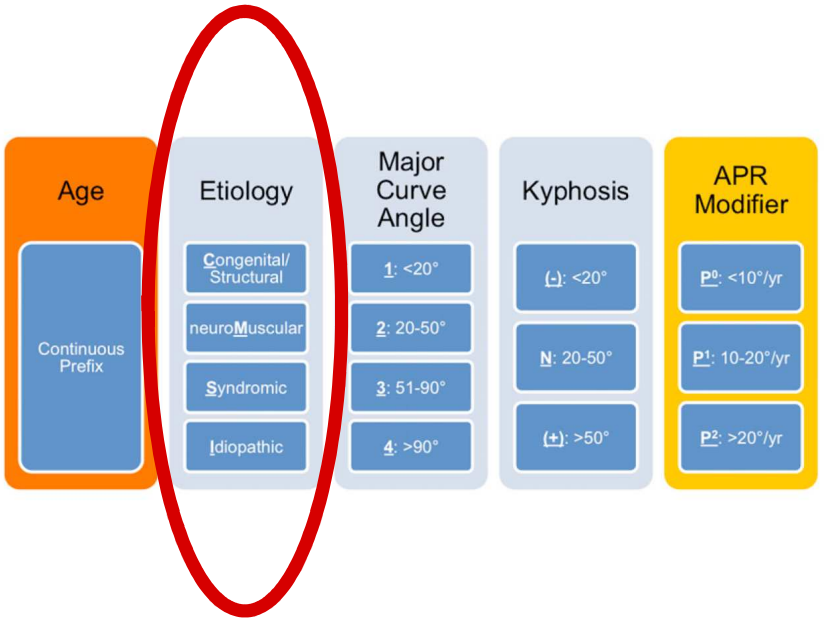
Conclusions

- EOSQ demonstrated known group validity when assessed on ~600 patients at multiple centers without the bias of treatment.
- Congenital patients scored (surprisingly?) well compared to Idiopathic patients (disease burden of congenital fusions not controlled)
- C-EOS Neuromuscular and Syndromic patients score significantly lower than Idiopathic and Congenital patients on most domains



Conclusions

- Future studies reporting EOSQ results of treatment must account for the C-EOS designations as matched or isolated groups
- Age and Cobb angle also have associations with EOSQ domain scores, albeit *weak* associations



Thank You

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