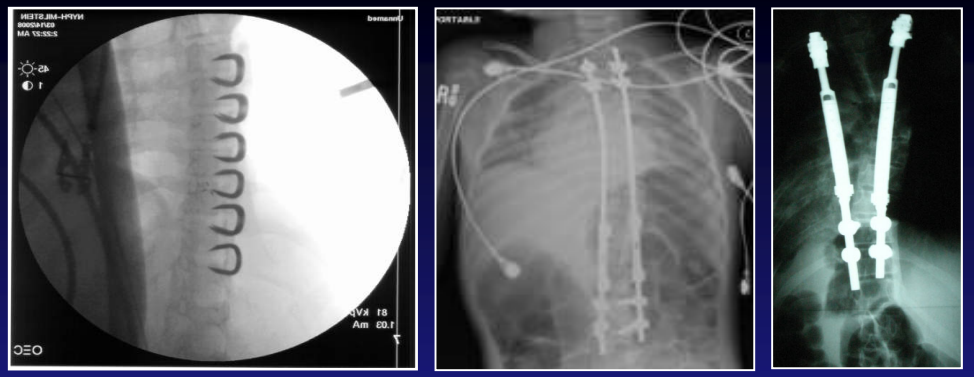

Health Related Quality of Life in Children with Early Onset Scoliosis:
Validation of a Disease Specific Instrument

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Background: Outcomes in EOS

- Improve Natural History
  - Xrays
  - Pulmonary function
- Improve Quality of Life
  - Patients with EOS can have significant perturbations in health related quality of life

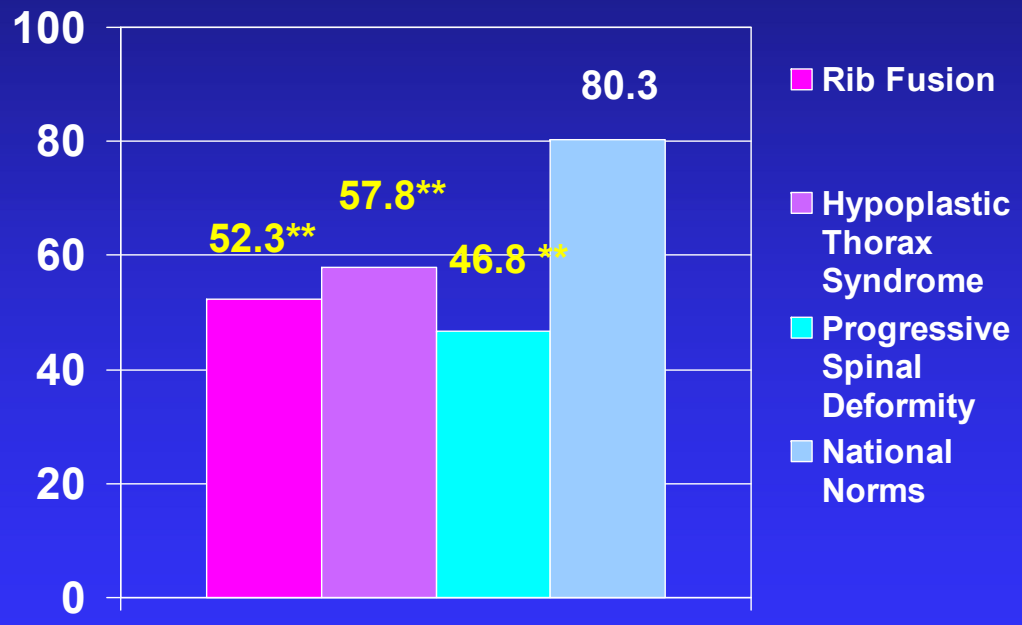
Retrospective Cohort Study of Pulmonary Function, Radiographic Measures and Quality of Life in Children with Congenital Scoliosis: An Evaluation of Patient Outcomes after Early Fusion

Vitale et al, 2006

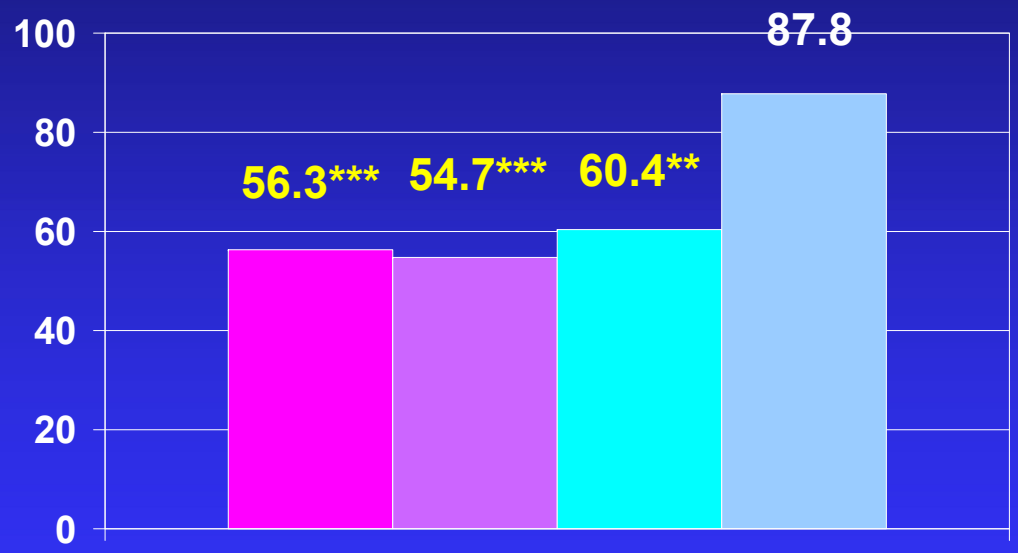
- **7 year follow up on 27 patients with early fusion (6 yrs avg) for congenital scoliosis**
- **Poor PFT, Poor QOL**
- ***Age at fusion* and *residual curve at follow up* seems to strongly drive lung function**
- **Quality of life appears to be largely independent of Cobb, age and other factors**

The Impact of Caring For a Child with TIS on the Family is Profound

CHQ in EOS; Vitale et al; JPO



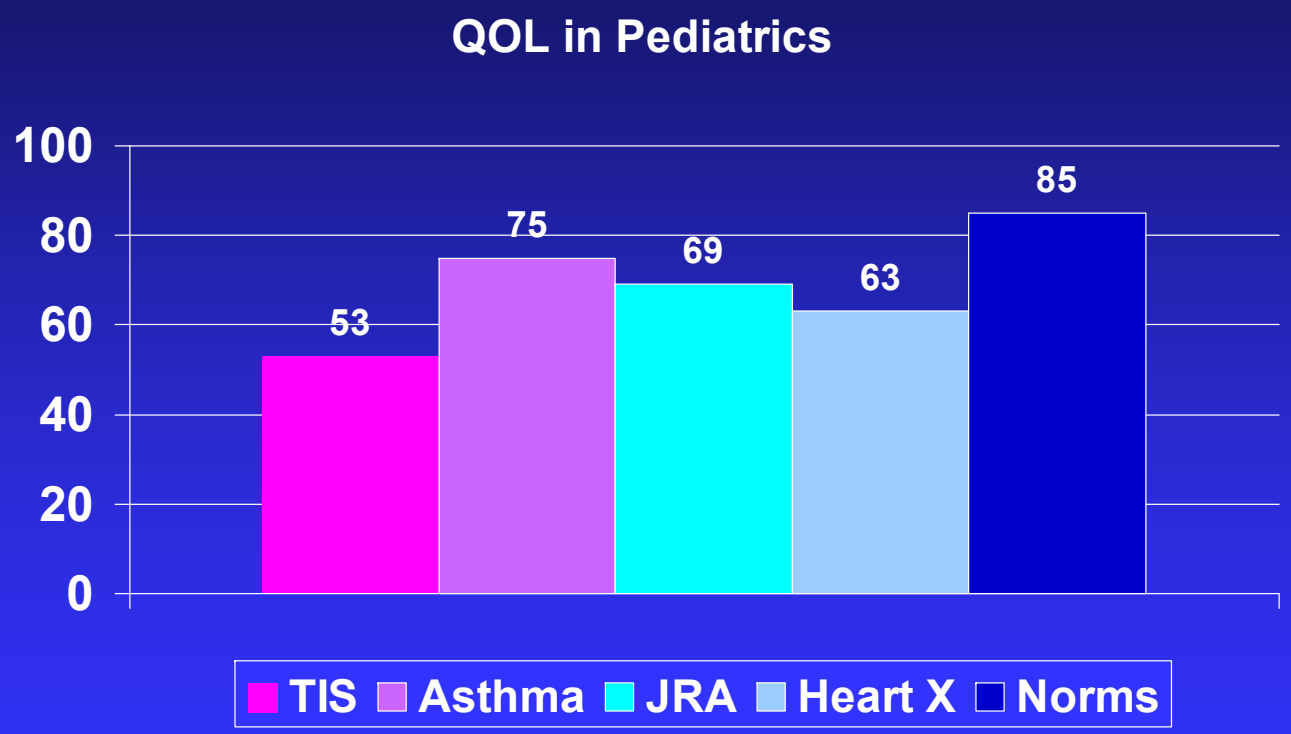
Parental Impact -Emotional



Parental Impact-Time

Conclusions: QOL (CHQ) in TIS

- These scores are among the lowest observed in pediatrics
  - Asthma
  - JRA
  - Heart transplant



Towards a Disease Specific Measure

- CHQ not public domain
- CHQ only for use in children >5 yo
- CHQ doesn't reflect specific issues with EOS
- Concerns about responsiveness
- You wouldn't use a thermometer to measure blood pressure !

Quality of life in EOS

- Hard thing to measure
  - Heterogenous population
  - Significant comorbidities
  - Age
  - Natural History may be subclinical in childhood

Background: Early Onset Scoliosis

Purpose: to develop a disease specific instrument (DSI) which reflects issues of importance to patients with EOS and caretakers, and is responsive to clinical changes after treatments.

Methods: Semi-Structured Qualitative Interview

- Questions were developed based on:
  - Analysis of CHQ items (Vitale, 2008)
  - Published patient-based HRQOL measures (ex. PedsQOL, SRS-30, PODCI, PEDI, SAQ)
  - Published pulmonary function. (ex. CRQ-SAS, PRQLQ, SGRG, ITG-CASF)

Methods: Semi-Structured Qualitative Interview

- Interviews were conducted on parents of children with a diagnosis of EOS undergoing surgeries
- Interviews conducted and transcribed

Methods: Master List of Items

- Qualitative interviews were interpreted using Framework technique
- 11 domains and 75 items were produced
  - General Health, Pulmonary Function, Physical Function, Transfer, Daily Living, Pain, Fatigue, Child Emotion, Surgical Burden, Parental Burden. Financial Burden

Methods: Validation

Content Validity

- Examined relativity and clarity of questions by 14 physicians, 3 health care providers, and 5 parents.
- Content validity indices (CVIs) were calculated.

Methods: Validation

Item Statistics

- Item Distribution
  - Ceiling and Flooring Effect (<80%)
  - Mean (3.0 for 5 point Likert scale)
  - Normal Distribution
- Item Reliability
  - Median Item Correlation (r= 0.4-0.5 between items within domains)

Patient Demographics

Gender	Male 36% Female 64%
Race	White (72.7%) Hispanic (18.2%) Other (9.1%)
Cobb Angle	45 degrees +/- 24 (20-100)
Mean age of child	5.9 +/- 2.4
Mean age of Primary Caregiver	39 (31-50)

Broad Range of Etiologies of EOS

Congenital	**(10/28) = 35.7%** 3 =Hemivertebrae and Fused Ribs 3 = No Fused Ribs 4 = T/ TL hemivertebrae
Neuromuscular	**(9/28) = 32.1%** 3 = SMA 2 = Mitochondrial Myopathy 1 = Congenital Muscular Dystrophy 1 = Merosin Deficient Muscular Dystrophy 2 = Marden Walker Syndrome (1 w/ tethered cord)
Spinal Cord	**(5/28) = 17.9%** 3 = Tethered cord (2 w/ Aicardi Syndrome) 1 = Myelomeningocele 1 = Chiari type I
Idiopathic	**(4/28) = 14.3%**

Surgical Treatment Plans

Preop/Observation		**11 (50%)**
Surgical Treatment		**11 (50%)**
	Limited Spinal Fusion	**4 (36%)**
	VEPTR	**4 (36%)**
	Growing Rod	**2 (19%)**
	Shilla Procedure	**1 (9%)**

Methods: Validation

Item Statistics

- **Item Validity**
  - **Item Total-Item correlation (r=0.3-0.5 when a domain has more than three items. Correlation in 1+2 and 3.)**
  - **Domains with more than three items after Item Distribution & Reliability and Contend Validity testing:**
    - **Parental Burden**
    - **Surgery**
    - **Treatment Outcome**

Methods: Psychometric Validation

Eg Activity of Daily Living Domain

Item #	Floor	Ceiling	Mean	Normal Distribution	Item Reliability	Relativity
33	20%	50%	3.40		.667 (vs. 34) .871 (vs. 35) .845 (vs. 36) .579 (vs. 37)	3.36
34	9.1%	81.8%	4.45		.667 (vs. 33) .795 (vs. 35) .677 (vs. 36) .767 (vs. 37)	3.35
35	10.0%	60.0%	3.90		.871 (vs. 33) .795 (vs. 34) .929 (vs. 36) .538 (vs. 37)	4.00
36	30.0%	60.0%	3.60		.845 (vs. 33) .677 (vs. 34) .929(vs. 35) .388 (vs. 37)	3.87
37	9.1%	72.7%	4.55		.579 (vs. 33) .767 (vs. 34) .538 (vs. 35) .388 (vs. 36)	4.30

Methods: Item Elimination

eg. parental burden

Item #	Corrected Item-Total Correlation
53	.736
54	.856
58	.790

Item #	Corrected Item-Total Correlation
53	.645
58	.645

Methods: Item Validity

Concerns about surgery

Item #	Corrected Item-Total Correlation
64	.643
65	.851
66	.057

Item #	Corrected Item-Total Correlation
64	.797
65	.797

Item does not belong to the "Surgery" domain

EOS DSI: 12 Domains and 24 Questions

- General Health
- Pain
- Pulmonary Function
- Physical Function
- Daily Living
- Fatigue
- Emotion
- Parental Burden
- Financial Burden
- Surgery
- Satisfaction
- Treatment Outcome

EOS DSI

OFFICE USE ONLY **Study ID:** **Date:** / /

General Health				
1. In general, you would say your child's health has been:				
Poor	Fair	Good	Very good	Excellent
2. How often has your child missed school due to his/her health condition?				
All of the time	Most of the time	Some of the time	A small amount of time	None of the time

Pain/Discomfort				
3. How often has your child had pain/discomfort?				
All of the time	Most of the time	Some of the time	A small amount of time	None of the time
4. How often did pain/discomfort interfere with your child's physical activities?				
All of the time	Most of the time	Some of the time	A small amount of time	None of the time
5. How often did pain/discomfort interfere with your child's sleep?				
All of the time	Most of the time	Some of the time	A small amount of time	None of the time

Pulmonary function

Next Steps

- Prospective Validation
- Testing of Responsiveness
- Develop Age-based Norms
- **Happy to share: email mgv1@columbia.edu**

Thank You

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