The CDH Study Group –
A possible model for multi-disciplinary collaboration?

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H1088 Joint Program
Section on Neonatal-Perinatal Medicine & Section on Surgery
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Faculty disclosure information

In the past 12 months, Krisa Van Meurs had the following financial relationship with the manufacturer of a commercial product discussed in this CME activity:

Ikaria/Mallinckrodt Pharmaceuticals - Advisory Panel

It is my obligation to disclose to you that I have served on an Advisory Panel for Ikaria. However, I acknowledge that today’s activity is certified for CME credit and thus cannot be promotional. I will give a balanced presentation using the best available evidence to support my conclusions and recommendations.

I do not intend to discuss an unapproved/investigative use of a commercial product/device in my presentation.
Presentation outline

- Why the CDH Registry was formed
- How the Registry is organized
- Pros and cons of the Registry
- What we have learned
- “Evidence based” best practices
- Using the CDH Registry for QI
- Moving forward
CDH Pathophysiology

Pulmonary hypoplasia

Remodeled pulmonary arteries

Intracardiac and extracardiac right to left shunts

Right ventricular systolic and diastolic dysfunction (pulmonary arterial hypertension)

Pulmonary arterial hypertension

Pulmonary venous hypertension

Left ventricular “hypoplasia” and dysfunction

Created by Satyan Lakshimrusimha, MD
Challenges to improving survival and outcomes

- With 1-4 cases per 10,000 births, CDH is officially a rare disease
- The degree of lung hypoplasia and pulmonary hypertension are variable making comparison of results problematic
- Few randomized clinical trials have been performed in the CDH population
- Management varies significantly across centers
Emergency Operation (Standard of Care)
Hyperventilation (Standard of Care)
HFOV (Unavailable)
ECMO (Rescue Therapy available at 20 US centers)
Role of the Heart (Unknown)
Surfactant (Untested)
Fetal Surgery (Unavailable)
Pulmonary Hypertension (Tolazoline)
Long Term Follow Up (Not Needed)
Overall Survival – 50%
Charter ELSO Meeting - 1989

EXTRACORPOREAL LIFE SUPPORT ORGANIZATION
Charter Meeting

October 1-3, 1989 Ann Arbor, Michigan
Members in attendance:

<table>
<thead>
<tr>
<th>NAME</th>
<th>CENTER</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lucienne Sanchez</td>
<td>CNMC, Washington, D.C.</td>
</tr>
<tr>
<td>Kevin Lally</td>
<td>Hermann Children's/Houston</td>
</tr>
<tr>
<td>Jim Atkinson</td>
<td>CHLA/Los Angeles</td>
</tr>
<tr>
<td>Charles Breaux, Jr.</td>
<td>Children's of Alabama</td>
</tr>
<tr>
<td>Karen West</td>
<td>Riley Hospital/Indpls IN</td>
</tr>
<tr>
<td>Billie Lou Short</td>
<td>CNMC, Washington, D.C.</td>
</tr>
<tr>
<td>William Engle</td>
<td>Riley Hospital/Indpls IN</td>
</tr>
<tr>
<td>Gail Kernaghan for W.P. Kanto</td>
<td>Med College of Georgia</td>
</tr>
<tr>
<td>Michele Walsh-Sukys</td>
<td>Rainbow Babies, Cleveland</td>
</tr>
<tr>
<td>David P. Meagher, Jr.</td>
<td>Children's Hospital, Denver</td>
</tr>
<tr>
<td>Gerald M. Haase</td>
<td>Children's Hospital, Denver</td>
</tr>
<tr>
<td>Jay Wilson</td>
<td>Boston Children's Hospital For Sick Children</td>
</tr>
<tr>
<td>Desmond Bohn</td>
<td>Johns Hopkins Hospital</td>
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<tr>
<td>Kyle Walker</td>
<td>Children's Hospital/Seattle</td>
</tr>
<tr>
<td>P. Pearl O'Rourke</td>
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</tbody>
</table>
The meeting was scheduled to begin at 15:30, and began shortly thereafter. It lasted for approximately one hour. Items of import discussed were as follows:

* There was universal agreement of a need for such a study group. The goals of the group were not completely defined, however 2 main goals were cited:

1) Universal data collection of CDH patients.

2) Collective attempt to answer questions regarding CDH patients. There was universal agreement that no single individual or institution had found "the answer" to the ubiquitous problem of CDH infants. There were numerous expressions of willingness to work together and attempt to put aside previous biases and large egos to collectively address CDH patient management and outcome.
CDH Study Group

- Dr. Kevin Lally at University of Texas Houston directs the Registry and Dr. Pam Lally is the data manager
- Data collection began in 1995, now over 9,000 patients
- Voluntary, IRB approved, no consent, limited data set with DOB and DOS
- Data available to CDH SG centers for analysis with minimum of 5 years participation
CDH Study Group Active Centers

- Currently, 69 active centers from 14 countries
- 36 centers have actively contributed since 1995
- Center volumes vary widely from 1-2 cases/year to >10 cases/year
The CDH Study Registry

Pros

- Ability to study infrequent problems
- Data on very large number of patients
- Individual centers can compare themselves with others
- Demonstrate changes over time with both management and outcome
The CDH Study Registry

Cons

- Observational data
- Inability to evaluate long-term sequelae
- Difficult to collect complicated information
- Wide spectrum of patients and treatment philosophies
| Versions of the CDH Database | 1995-2000 | medications
ventilation strategies
ECMO |
|-----------------------------|-----------|------------------------------------------------|
| 2001-2005                   | method of delivery
O2/CO2 values
discharge information (NG feeds, oxygen, meds)
associated cardiac anomalies |
| 2006-2014                   | defect size and related outcomes
reasons for non-repair
PH severity and management |
| 2015- present               | prenatal US and MRI
echocardiograms |
What have we learned?

- Demographics of CDH population over time
- Longitudinal changes in treatments used
- Do specific treatments or management improve outcome
- Center differences
- Staging
## Changes in demographics and outcomes

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<td>Prenatal diagnosis (%)</td>
<td>47</td>
<td>56</td>
<td>65</td>
<td>69</td>
</tr>
<tr>
<td>Inborn (%)</td>
<td>34</td>
<td>39</td>
<td>44</td>
<td>49</td>
</tr>
<tr>
<td>Left Side (%)</td>
<td>80</td>
<td>82</td>
<td>84</td>
<td>84</td>
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<tr>
<td>Isolated (%)</td>
<td>86</td>
<td>89</td>
<td>89</td>
<td>87</td>
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<tr>
<td>ECMO (%)</td>
<td>36</td>
<td>30</td>
<td>28</td>
<td>30</td>
</tr>
<tr>
<td>Survival (%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>All CDH</td>
<td>68</td>
<td>69</td>
<td>70</td>
<td>71</td>
</tr>
<tr>
<td>Isolated CDH</td>
<td>72</td>
<td>73</td>
<td>74</td>
<td>76</td>
</tr>
<tr>
<td>ECMO CDH</td>
<td>53</td>
<td>51</td>
<td>49</td>
<td>52</td>
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</tbody>
</table>

**Conclusions:**

- Prenatal diagnosis continues to increase; however, many are still transferred for surgery or ECMO
- Survival rates are slowly increasing
- ECMO use has declined over time
Longitudinal changes in treatment strategies
Operative Approach 1995-1996

- Subcostal: 90%
- Thoracic: 6%
- Other: 4%
Operative Approach 2014

- Subcostal: 71%
- Thoracoscopic: 15%
- Thoracic: 4%
- Other: 2%
- Laparoscopic: 6%
Timing of CDH Repair 1995-1996

- 0-24 hours: 35%
- 24-48 hours: 15%
- 48-72 hours: 10%
- 72-96 hours: 10%
- 96-120 hours: 5%
- 120-240 hours: 10%
- >240 hours: 5%
Timing of CDH Repair 2011-2012

Hours of Age

- 0-24
- 24-48
- 48-72
- 72-96
- 96-120
- 120-240
- >240

%
% Patients with ECMO After Repair
Specific treatment strategies and outcome
ORIGINAL ARTICLE

Does a highest pre-ductal O₂ saturation <85% predict non-survival for congenital diaphragmatic hernia?

BA Yoder¹, PA Lally² and KP Lally², The Congenital Diaphragmatic Hernia Study Group
Prediction of survival using best pre-ductal saturation, PaO$_2$ and PaCO$_2$

**Design:** Analysis of highest pre-ductal sat, highest PaO$_2$, lowest PaCO$_2$ in first 24 hours. Major anomalies were excluded.

**Results:**


N=3816
Prediction of survival using best pre-ductal saturation, PaO2 and PaCO2

Results:

- No identifiable cut-off value using pre-ductal saturation, PaO2, or PaCO2 could be found that achieved a high positive or negative predictive value for survival.
- Survival was 44% for infants with pre-ductal O2 sat <85% who were repaired.
- 83% of the survivors required ECMO.

Conclusion:

- Pre-ductal saturation <85% is not uniformly fatal
- Limiting interventions in CDH may result in unnecessary deaths.

Does ECMO improve survival in newborns with CDH?

**Design:**
- ECMO benefit assessed by comparing survival in newborns with similar mortality risk using validated predictors, birth weight and 5 minute Apgar.

**Results:**
- Overall survival in non-ECMO group 77% vs 52% ECMO group
- After categorization into risk quintiles, ECMO improves survival only for those with mortality risk >80%

![Graph showing survival rates across different risk quintiles](image)

**Predictive mortality risk (%)**
- N=632

Outcomes in surfactant and non-surfactant treated CDH infants

**Conclusion:** Analysis of observational data fails to identify a benefit associated with surfactant therapy in the term infant with CDH

Does timing of repair on ECMO impact outcome?

Design: Retrospective analysis of CDH SG Registry from 1995-2005 including 636 infants who received ECMO and repair

Results: 

Conclusions: CDH repair after ECMO is associated with improved survival compared to repair on ECMO.

Does minimally invasive repair impact re-herniation rates?

**Design:**
Using CDH SG Registry data from 1995-2010, hernia recurrence rates were compared in open versus minimally invasive cases.

**Results:**
Open repair had re-herniation rate 2.7% compared to 7.9% in MI group, OR 3.59 (CI 1.92-6.71) with adjustment for 4 factors (GA, BW, patch repair, ECMO)
Re-herniation higher with both primary repair and patch repair in MI group (6.1% and 8.8%) while low in primary and patch repair in Open group (3.8% and 1.6%)

**Conclusions:**
MI repair is associated with higher risk of re-herniation

Center differences
Center differences

- Participating centers who are current
- At least 10 cases/year (14 centers)
- Evaluated survival, defect size and ECMO use
## Results

### Center Characteristics

<table>
<thead>
<tr>
<th>Centers</th>
<th>% Repaired</th>
</tr>
</thead>
<tbody>
<tr>
<td>Overall *</td>
<td>81% 83% 84% 92% 84% 90% 81% 86% 81% 75% 69% 81% 80%</td>
</tr>
</tbody>
</table>

*Centers (range 69%-92%, *p<0.05)*
Results

Center Characteristics

% Prenatally Diagnosed

Centers
(range 41% - 88%, * p< 0.05)
Results

Center Characteristics

% Patch Repair

(range 25%-67%, *p<0.05)
Results

Center Characteristics

% Agenesis

Centers
(range 3%-29%, *p<0.05)
# Results

## Center Characteristics

<table>
<thead>
<tr>
<th>Center</th>
<th>Survival</th>
<th>Birth Weight (kg)</th>
<th>EGA (weeks)</th>
<th>1-minute Apgar</th>
<th>5-minute Apgar</th>
<th>Associated Anomalies</th>
<th>Defect Characteristics</th>
<th>Treatment Characteristics</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Cardiac</td>
<td>Left-sided CDH</td>
<td>Prenatally Diagnosed</td>
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<tr>
<td>2</td>
<td>77%</td>
<td>3.1</td>
<td>38.2</td>
<td>5.3</td>
<td>7.0</td>
<td>6%</td>
<td>86%</td>
<td>58%</td>
</tr>
<tr>
<td>4</td>
<td>77%</td>
<td>3.0</td>
<td>37.7</td>
<td>4.7</td>
<td>6.7</td>
<td>10%</td>
<td>78%</td>
<td>70%</td>
</tr>
</tbody>
</table>

|        |          |                   |            |                |               | Chromosomal          | Agenesis              | Inborn                  |
|        |          |                   |            |                |               |                      |                       | ECMO                     |
|        |          |                   |            |                |               |                      |                       | Repaired                 |
|        |          |                   |            |                |               |                      |                       | 84%                     |

p < 0.05
## Results

### Center Characteristics

<table>
<thead>
<tr>
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<th>Birth Weight (kg)</th>
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<th>5-minute Apgar</th>
<th>Associated Anomalies</th>
<th>Defect Characteristics</th>
<th>Treatment Characteristics</th>
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<tr>
<td>3</td>
<td>61%</td>
<td>3.0</td>
<td>38.2</td>
<td>5.2</td>
<td>6.7</td>
<td>4% 5%</td>
<td>80% 9%</td>
<td>58% 60% 46% 72%</td>
</tr>
<tr>
<td>12</td>
<td>60%</td>
<td>2.9</td>
<td>37.5</td>
<td>5.0</td>
<td>7.3</td>
<td>8% 2%</td>
<td>83% 8%</td>
<td>88% 1% 0% 69%</td>
</tr>
</tbody>
</table>

\[ p < 0.05 \]
Center differences

- Very wide disparities in risk distribution
- Very wide disparities in management
- Very wide disparities in surgical treatment
- Reporting needs to be standardized
Risk-stratified outcomes

- Wide variation in patient characteristics amongst “high volume” centers
- Wide variation in therapies among centers
- Centers with similar outcomes may have heterogeneous patient and treatment profiles
- Risk adjustment critical
- Standardized reporting will be important to evaluate therapies
From research to QI

- What is the role of ECMO in CDH?
- Who has the “Best Outcomes” for CDH?
- What are the Best Treatments?
- Can/should we standardize care?
- Can/should we regionalize care?
Is ECMO Beneficial in Infants with CDH?
TRADITION

Just Because You’ve Always Done It That Way
Doesn’t Mean it’s Not Incredibly Stupid.
ECMO in CDH: Current selection criteria

- Oxygenation Index
- NPI II
- AaDO2 > ? X ? Hours
- paO2 < ? X ? Hours
- Nucleated RBC
- Clinical judgment
- Failure to “Stay in the Box”
% Patients with ECMO After Repair

Year

%
ECMO use – Top 25 centers by volume
Centers with >60% Stage III-V and NR survival ECMO use
Centers with >60% Stage III-V and NR survival ECMO use
Is ECMO Beneficial in Infants with CDH?

Maybe?
What is the “Best Practice” for CDH?
What is the “Best Practice” for CDH?

We should use the best possible evidence.
CDH Best Practice

Medline – Congenital Diaphragmatic Hernia

Randomized Clinical Trials

- 29 Citations
- iNO – 10 (40%)
- ECMO – 5 (4 UK Trial)
- Tracheal Occlusion 4 (3 UCSF, 1 Ruano)
- 2 Delayed OR
CDH Best Practice

Medline – Congenital Diaphragmatic Hernia

Randomized Clinical Trials

4 since 2006
CDH Best Practice

Medline – Congenital Diaphragmatic Hernia

Randomized Clinical Trials

There have been ZERO appropriately designed and powered studies published in CDH
CDH Best Practice

- VICI Trial – 31 Individual Points to Protocol
  - Level of Evidence – All Grade “D”
CDH Best Practice

MANAGEMENT OF INFANTS WITH CONGENITAL DIAPHRAGMATIC HERNIA FROM BIRTH TO SURGERY
Amir M. Khan and Kevin P Lally
(Copyright 2011-2013: The University of Texas Health Science Center at Houston, Department of Pediatrics, Division of Neonatal-Perinatal Medicine)

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<thead>
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<tbody>
<tr>
<td>[A]</td>
<td>Large randomized trial(s)</td>
<td>[E]</td>
<td>Expert opinion or consensus</td>
</tr>
<tr>
<td>[B]</td>
<td>Small randomized trial</td>
<td>[F]</td>
<td>Basic laboratory research</td>
</tr>
<tr>
<td>[C]</td>
<td>Prospective cohort study</td>
<td>[L]</td>
<td>Legal requirement</td>
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<tr>
<td><strong>[D]</strong></td>
<td>Retrospective cohort or case control study</td>
<td>[Q]</td>
<td>Decision Analysis</td>
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<tr>
<td>[S]</td>
<td>Review article</td>
<td>[X]</td>
<td>No evidence</td>
</tr>
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36 Different Points in the analysis
1 Level “D”
35 Level “E”
Can We Standardize Care for CDH?

Outcome in Infants with Congenital Diaphragmatic Hernia: The Role of a Standardized Postnatal Treatment Protocol

Using the CDHR for Quality Improvement
IDIocy

Never Underestimate the Power of Stupid People in Large Groups.
Comparing outcomes
Step 1
Comparing outcomes

Step 1

Talk the Same Language
Comparing outcomes
Standardized reporting for congenital diaphragmatic hernia – An international consensus

Methods: Factors Evaluated

- Defect class
- Cardiac anomalies
- Chromosomal anomalies
- Birthweight /Gestational age
- Apgar Scores
Grading defect size

A
B
C
D
# Results

<table>
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<tr>
<th>Defect</th>
<th>Total</th>
<th>Survived</th>
<th>% Survival</th>
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<tbody>
<tr>
<td>A</td>
<td>164</td>
<td>163</td>
<td>99</td>
</tr>
<tr>
<td>A+</td>
<td>8</td>
<td>7</td>
<td>88</td>
</tr>
<tr>
<td>B</td>
<td>572</td>
<td>551</td>
<td>96</td>
</tr>
<tr>
<td>B+</td>
<td>18</td>
<td>12</td>
<td>67</td>
</tr>
<tr>
<td>C</td>
<td>372</td>
<td>291</td>
<td>78</td>
</tr>
<tr>
<td>C+</td>
<td>27</td>
<td>15</td>
<td>56</td>
</tr>
<tr>
<td>D</td>
<td>144</td>
<td>84</td>
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<td>D+</td>
<td>18</td>
<td>7</td>
<td>39</td>
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<tr>
<td>NR</td>
<td>326</td>
<td>1</td>
<td>&lt;1</td>
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## Results

<table>
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<th>Defect</th>
<th>Total</th>
<th>Survived</th>
<th>% Survival</th>
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<tbody>
<tr>
<td>A</td>
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<td>II</td>
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<tr>
<td>III</td>
</tr>
<tr>
<td>IV</td>
</tr>
<tr>
<td>V</td>
</tr>
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</table>
Survival by stage
CDH classification

- Patients with CDH can be placed into easily characterized groups.
- Future reporting should include the stage as well as the number of “Non-Repair” patients
- Focus on Stages III-V and Non-Repair
Comparing outcomes
Step 2
Comparing outcomes

Step 2

Where Am I?
<table>
<thead>
<tr>
<th>Defect Type</th>
<th>Your Center</th>
<th></th>
<th></th>
<th></th>
<th></th>
<th>The CDH Registry</th>
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<tr>
<td></td>
<td>N</td>
<td>N Surv</td>
<td>% Surv</td>
<td>% Distrib*</td>
<td>N</td>
<td>N Surv</td>
<td>% Surv</td>
<td>% Distrib*</td>
<td></td>
</tr>
<tr>
<td>A</td>
<td>8</td>
<td>8</td>
<td>100%</td>
<td>9%</td>
<td>436</td>
<td>430</td>
<td>99%</td>
<td>11%</td>
<td></td>
</tr>
<tr>
<td>B</td>
<td>28</td>
<td>26</td>
<td>93%</td>
<td>32%</td>
<td>1244</td>
<td>1177</td>
<td>95%</td>
<td>31%</td>
<td></td>
</tr>
<tr>
<td>C</td>
<td>35</td>
<td>28</td>
<td>80%</td>
<td>40%</td>
<td>1047</td>
<td>827</td>
<td>79%</td>
<td>26%</td>
<td></td>
</tr>
<tr>
<td>D</td>
<td>9</td>
<td>4</td>
<td>44%</td>
<td>10%</td>
<td>421</td>
<td>239</td>
<td>57%</td>
<td>11%</td>
<td></td>
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<tr>
<td>No Repair</td>
<td>7</td>
<td>0</td>
<td>0%</td>
<td>8%</td>
<td>637</td>
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<tr>
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<td>0</td>
<td>0</td>
<td>0%</td>
<td>8%</td>
<td>98</td>
<td>92</td>
<td>94%</td>
<td>2%</td>
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<tr>
<td>High Risk</td>
<td>51</td>
<td>32</td>
<td>63%</td>
<td>59%</td>
<td>2105</td>
<td>1078</td>
<td>51%</td>
<td>53%</td>
<td></td>
</tr>
<tr>
<td>(C + D + Non Repair)</td>
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<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Your Center</td>
<td>The CDH Registry</td>
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<td><strong>Survival</strong></td>
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<td>All CDH</td>
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<td><strong>Survivors at 30 days of Life</strong></td>
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<td>On O2</td>
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<td>On Room Air</td>
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<td>1592</td>
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Comparing outcomes
Step 3: Implementation of change
The Future of CDH – Moving Forward

Observational studies
- RCTs very, very unlikely (NEST)—cost, sample size, ...
The Future of CDH – Moving Forward

Observational studies
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Novel statistical designs
- Expanded involvement in registries can lead to quicker analyses of changes
The Future of CDH – Moving Forward

Observational studies
- RCTs very, very unlikely (NEST)—cost, sample size, ...

Novel statistical designs
- Expanded involvement in registries can lead to quicker analyses of changes

Link to quality/risk adjusted outcomes
- Need structure in place to achieve this
Moving Forward

- Networks
- Should we Regionalize Care?
- Coordinated Follow-up
- Cost
Regionalizing CDH care

Impact of hospital volume on in-hospital mortality of infants undergoing repair of congenital diaphragmatic hernia

Regionalizing CDH care

Survival Rate in Congenital Diaphragmatic Hernia: The Experience of The Canadian Neonatal Network

By Patrick J. Javid, Tom Jaksic, Erik D. Skarsgard, Shoo Lee, and the Canadian Neonatal Network

Boston, Massachusetts and Vancouver, British Columbia

JPS 2004
Regionalizing CDH care

Effect of hospital case volume on outcome in congenital diaphragmatic hernia: the experience of the Canadian Pediatric Surgery Network

Jeremy R. Grushka\textsuperscript{a}, Jean-Martin Laberge\textsuperscript{a,*}, Pramod Puligandla\textsuperscript{a}, Erik D. Skarsgard\textsuperscript{b}
the Canadian Pediatric Surgery Network

JPS 2009
Regionalizing CDH care

Box plot showing O/E mortality ratio by center group

Lo: 1.21
HiHi: 0.94
HiLo: 0.81

p<0.0001, Tukey
Regionalizing CDH care

- All published studies show volume benefit
- Currently no stimulus ($) to change
- Many factors at play
Conclusions

- What about cost?
- What are the best outcomes to measure?
  Survival, 18 month NDI, School age, Workforce, ???
- Who is going to do this?
- How are we going to do this?
Conclusions

- What about cost?
- What are the best outcomes to measure?
  Survival, 18 month NDI, School age, Workforce, ???
- Who is going to do this?
- How are we going to do this?

I HAVE NO IDEA
Final Advice

The secret of enjoying a good wine:

1. Open the bottle to allow it to breathe.

2. If it does not look like it’s breathing, give it mouth-to-mouth.
more information on this subject, see the following publications:

