MAKING THE GRADE:
INVESTIGATING ACADEMIC OUTCOMES OF CHILDREN WITH CLEFT LIP AND PALATE

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• I do not intend to discuss an unapproved/investigative use of commercial products/devices.
THANK YOU CINDY
OBJECTIVES

• Illustrate trends in academic outcomes for children with orofacial clefts
• Evaluate potential factors that contribute to observed academic deficits
• Propose an intervention to improve outcomes
OROFACIAL CLEFTS: 
THE BASICS

- CLP: 1 in 700 births
- CP: 1 in 2000 births
- Genetic and environmental factors
- Isolated vs Syndromic

Functions of the palate

- FEEDING
- HEARING
- SPEECH
TIMELINE OF CARE

**Newborn visit**
- Lip repair
- Palate repair

**Cleft repair**
- Ear tubes placed

**Hearing screens**
(birth, 9mo, & annually)

**Speech therapy** (if needed)

**Speech surgery** (if needed)

**Lip/nose revision (if needed)**

**Bone graft (if needed)**

**Lip/nose revision, maxillary advancement (if needed)**

**Orthodontics: phase 1**

**Orthodontics: phase 2**

**Dental care**
PRENATAL DIAGNOSIS

Will I feel comfortable taking my baby in public before lip repair?

Will my baby have other problems?

Will other people bully my child because of the cleft?

Is this because of something I did wrong?

Does this mean my child will have learning problems?
IMPACTS OF OROFACIAL CLEFTS

“There appears to be an innate human tendency to associate craniofacial malformations with abnormal cognitive development.”

Cunningham, 2007
Key Point:
This does not mean that every child with an orofacial cleft will have academic deficits.
Systematic Review

- Search strategy
  - Medline, Embase, PsychInfo, CINAHL
  - 1980-2017

- Search terms
  - Cleft palate, cleft lip, orofacial cleft
  - Terms to include neurodevelopmental and academic outcomes
Inclusion
- Patients <25 years with orofacial clefts
- Measures of neurodevelopmental outcomes
- English language
- Middle to high income economies

Exclusion
- <10 cases
- Qualitative studies
2270 references identified by search strategy
110 duplicate references excluded

2160 references screened for inclusion using title and abstract, with full-text consulted when necessary

2080 references excluded that did not meet inclusion criteria
80 references considered for inclusion by 2 authors (EG, BC)

31 references included in final analysis

52 papers excluded that did not meet inclusion criteria on closer review

3 additional references identified

Number of papers for each age group:
  - Infant/toddler: 10
  - Early school aged: 14
  - Adolescence: 7
Unique cohort and outcomes
Validated outcome measurement used
Homogeneous group studied
Control group included
Hearing data included
Clear identification of syndromic patients
Participation rate reported
Adequate demographic data reported
Population based sample

SYSTEMATIC REVIEW: QUALITY SCORES

Gallagher and Collett, Pediatrics, 2019
• Quality of studies was variable

• Several high quality studies clearly show academic deficits

• Deficits were present in a range of domains and ages

• Future studies should include more rigorous review of participants

• Children with orofacial clefts are at risk for neurodevelopmental deficits and should be monitored and supported
• Evaluating neurodevelopmental outcomes is complex

• Functional and psychosocial impacts of orofacial clefts

• Many potential factors
  • Intrinsic
  • Extrinsic
INTRINSIC DIFFERENCES

• Normally, adult gaze focuses on an infant’s eyes before 6 weeks, then includes more time on the mouth when infant starts to vocalize.
• Maternal eye contact predicts mother-infant relationship 1 year later and has been linked to developmental outcomes.
• Maternal gaze was shifted when infant had a cleft lip
  • Gaze towards infant’s body
  • Gaze towards facial areas other than eyes or mouth.

Does breastfeeding have a positive impact on cognition and behavior for children?

- Nutritional benefits support neural maturation and may impact language development
- Some studies found better neurodevelopmental outcomes after exclusive breast milk feeding
- More recent studies have been less clear
- Skin-to-skin contact may help with bonding and subsequently behavior

AAP policy statement, 2012
5 year retrospective review of children with cleft palate
Breast milk feeding (ever) was 29.5%
CDC report = 81%
Lower z-scores for weight and weight for length

Kaye et al. Initial Nutritional Assessment of Infants With Cleft Lip and/or Palate: Interventions and Return to Birth Weight; Cleft Palate-Craniofacial Journal, 2017.
VELO:
VPI Effects on Life Outcomes

- Speech Limitations
- Swallowing problems
- Situational difficulty
- Emotional impact
- Perception by others
- Caregiver impact
In non-cleft populations, few clear differences have been identified in developmental outcomes after anesthesia.

Danish study: neurodevelopmental outcomes of CL, CLP, CP
- CL had higher scores, CP lowest scores
- Cleft type, not number of surgeries, was associated with lower outcomes.

Sun et al. Association Between a Single General Anesthesia Exposure Before Age 36 Months and Neurocognitive Outcomes in Later Childhood. 2017, JAMA.
SCHOOL ABSENCE

- Higher absence rates for CLP in grades 4-6
- No difference in high school
- Higher absences associated with lower standardized test scores
- Children with CP had lower scores regardless of absence rates

Population-based cohort in Western Australia
400 cases, 1800 controls
Quantifying school absence for children with orofacial clefts
Impact of school absence on test scores

Bell et al., School Absence and Its Effect on School Performance for Children Born with Orofacial Clefts; Birth Defects Research, 2017.
Psychosocial Outcomes

• Narrative review of 148 quantitative and qualitative studies, 2004-2015

• 5 domains of adjustment:
  • Developmental trajectory
  • Behavior
  • Emotional Well-being
  • Social Experiences
  • Satisfaction with Appearance and Treatment

• Contradictory results in all areas but overall impact of cleft seems low

Psychological adjustment to cleft lip and/or palate: A narrative review of the literature
Nicola Marie Stock* and Kristin Billaud Feragen*

*Centre for Appearance Research, University of the West of England, Bristol, UK; Senter for sjeldne diagnoser (Centre for Rare Disorders), Oslo Universitetssykehus HF, Oslo, Norway
(Received 8 September 2015; accepted 12 January 2016)
HEARING LOSS

Conductive hearing loss
Chronic middle ear effusions
Chronic otitis media
Early Identification of Hearing Loss - Method

1:3:6

Screening
- No later than 1 month of age

Diagnostic testing with Pediatric Audiologist
- No later than 3 months of age

Early Intervention
- No later than 6 months of age
What is the degree of hearing loss before palate repair for infants with cleft palate?

- Retrospective chart review
- Cleft palate ± cleft lip
- DOB 2008-2015
- Palate repaired at SCH before age 3 years
- Audiograms in AudBase

Tubes placed with palatoplasty
DEGREE OF HEARING LOSS BY AGE

CLICK-EVOKED BAER, n=61
- 49.2%: mild hearing loss
- 11.5%: moderate or greater hearing loss

BEHAVIORAL AUDIOGRAMS, n=259
- 32.0%: mild hearing loss
- 30.5%: moderate or greater hearing loss

Gallagher et al. In progress.
HEARING AFTER PALATE REPAIR

4.6%: mild or greater hearing loss

Gallagher et al. In progress.
Potential targets for intervention:

- Parental bonding
- Breastmilk feeding
- Hearing loss
- Home language environment
INTERVENTION STUDY

Can we change the home language environment?

Oral Cleft

Home Language/Literacy Environment
- Shared Oral Reading
- Reciprocal Conversation
- Parent Beliefs about Reading/Development

Pre-Reading Skills
- Vocabulary/Grammar
- Print Awareness
- Phonological Awareness
Can Reach Out and Read be used to positively impact the Home Language Environment?

www.reachoutandread.org
EVIDENCE FOR REACH OUT AND READ

- Improves home literacy environment
  - Frequency of shared reading
  - Availability of books in the home
  - Reading becomes a favorite shared activity
- Increases scores on testing
  - Receptive and expressive language
  - Literacy scores at school entry
  - Low socioeconomic settings
  - English and non-English-speaking children
CRANIOFACIAL REACH OUT AND READ

- 2012: partnered with national ROR
- Developed a list of books by age and specific speech sounds
- Follow ROR model but also demonstrate how to use books to practice speech
MEASURING THE HOME LANGUAGE ENVIRONMENT

- 16 hours of recordings at home
- Sorts child vs others, TV, radio
- Software analyzes and provides
  - Adult Word Count
  - Child Vocalizations
  - Conversational Turns

LENA device
• Feasibility study
  • Recruitment
  • Protocol implementation

• Study population
  • Goal: 60 children with clefts
  • 9 months (±2 months)
LENA ROR STUDY

• Inclusion
  • CL, CLP, CP
  • SCH Craniofacial Center
  • English or Spanish-speaking

• Exclusion
  • Syndrome with known delays
  • Brain malformation, seizure
  • Profound hearing loss
  • Hypotonia
  • Hospitalized >6 weeks
  • State custody, adopted
STUDY DESIGN

Group 1

Group 2

Group 3

9 mos

18 mos

24 mos

[Diagram showing the study design with different groups and time points labeled.]
BASELINE CHARACTERISTICS OF PARTICIPANTS

- 78 approached, 27 enrolled
- Consent rate: 35%

- Combined recordings with others from a different study to increase pre-intervention recordings

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<th>N or mean</th>
<th>% or SD</th>
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<td>Age, months</td>
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<tr>
<td>Cleft type</td>
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<td>Cleft lip</td>
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<td>Cleft palate</td>
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<td>Cleft lip and palate</td>
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• Improvement in slope of the curve after intervention?
• Home language environment is a modifiable target of an intervention
• Feasibility study, need larger sample size
• Future plans: multicenter randomized trial with reading intervention and coaching
IMPROVING ACADEMIC OUTCOMES

MODIFIABLE TARGETS

- BONDING
- HEARING LOSS
- BREAST MILK
- HOME LANGUAGE

POTENTIAL INTERVENTIONS

- COACHING
- AMPLIFICATION
- PARENT SUPPORT
- READING
QUESTIONS?

• Thank you!
  • Craniofacial Center
  • Seattle Children’s Hospital Academic Enrichment Fund
  • Research collaborators
  • Patients and families