Toddlers with Cleft Palate: Understanding and Intervening to Address Differences in Speech and Language Development

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Disclosure Statement

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‣ An Exploratory Model of Early Speech Intervention for Children with Cleft Palate, (#1R21DC009654), 2009-2011, Nancy J. Scherer, Principal Investigator.
‣ There are no other conflicts of interest

Introduction and Overview

‣ Provide a general overview of the communication needs of toddlers with CLP
‣ Present 3 studies related to early speech and language intervention
   ◦ Assessment of Toddlers with CLP– Jennifer Frey
   ◦ Effects of Intervention: Language Outcomes – Ann Kaiser
   ◦ Effects of Intervention: Speech Outcomes– Nancy Scherer
‣ Discuss the needs for future research
‣ Discuss specific implications for practice
‣ Answer questions from the audience

Cleft Lip and/or Palate (CLP)

‣ 4th most common birth defect in the United States
   ◦ Affects an estimated 1 in 750 births
‣ Variable speech and language development in young children with CLP
   ◦ Children with CLP have more atypical patterns of articulation compared to age-matched children without CLP
‣ After repair, children with CLP have the capacity to produce normal speech
   · Persistent speech errors
   · Atypical articulation
   · Differences in use of language in naturalistic settings

Critical need for research

‣ Few studies describing early development of speech and language in children with CLP
‣ Very few studies of early intervention
‣ The description of outcomes for children with CLP are mixed
   ◦ Need for method appropriate for toddlers
   ◦ Need for precise description of speech and language development in first 3 years
   ◦ Population is heterogeneous and often not well-described
   ◦ No RCT of early interventions for toddlers

What are best practices for early intervention for toddlers with CLP?

‣ Advances in surgical and post surgical management
‣ Determining when both speech and language communication intervention is warranted (vs. waiting)?
‣ Sensitive, age appropriate strategies for assessment are needed
‣ Naturalistic interventions are developmentally appropriate but not yet well-researched
   ◦ Few studies of toddlers and preschoolers
   · Scherer, 1999
   Positive outcomes

Assessment of Speech and Language Skills of Toddlers With and Without Cleft Palate

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Study Purpose

- Describe and compare speech and language skills of young children with CLP to age and gender matched children without CLP
  - Across measures
  - Across materials
  - Across conversational partners

Research Questions

1. Do children with CLP differ from typically developing toddlers on standardized measures of language and speech?

1. Do children with CLP differ in their use of language in interactions with therapists and caregivers?

Participants

- 48 children between 13 and 37 months old
  - 24 children with nonsyndromic, repaired CLP
  - 24 children with typical speech and language development (TL)
    - age (± 1 month) and gender matched samples

	Children with CLP			Children with TL		
	Mean	**SD**	**Range**	**Mean**	**SD**	**Range**
Child Age (months)	26.00	6.01	(14 36)	25.71	6.34	(13, 37)
Caregiver Age (years)	32.23	5.76	(21, 43)	34.13	4.79	(25, 44)

Inclusion Criteria

- **All participants**
  - Cognitive scale composite score of 80 or higher on the Bayley-III
  - Could produce at least 5 different words per parent report
  - Hearing within normal range

- **Children with CLP**
  - Initial palate repairs prior to 12 months old
  - No syndrome diagnosis

Cognitive & Language Assessment

- **Standardized, norm-referenced assessment**
  - Bayley-3 Cognitive Subtest
  - PLS-4
    - Auditory Comprehension
    - Expressive Communication
    - Total Communication

- **Parent Report**
  - MCDI
    - Total words

- **Play-based language sample with clinician**

- **Play session with caregiver**

Measuring Use of Language

- **Language Sample & Play Session**
  - Total number of words spoken (TNW)
  - Number of different words spoken (NDW)
  - Mean length of utterance in morphemes (MLUm)
  - Intelligibility: % of intelligible utterances

Results: Standardized Assessment & Parent Report

|	Children with CLP			Children with TL			Effect Size
	Mean	SD	Range	Mean	SD	Range	d
Bayley Cognitive	99.8	10.4	(80, 120)	102.7	10.3	(90, 140)	−0.28
PLS AC	102.5	17.0	(67, 129)	108.8	15.0	(85, 136)	−0.39
PLS EC	102.1	15.9	(74, 128)	111.3	18.8	(85, 147)	−0.53
PLS TC	102.7	17.2	(68, 132)	111.2	17.8	(85, 146)	−0.49
MCDI total words	263.2	246	(6, 642)	343.3	217	(17, 671)	−0.35

- No statistically significant differences between groups on standardized measures or parent report
- Results from standardized measures indicate mean performance in each group fell in average range

Results: Use of Language in Play

Language Sample	Children with CLP			Children with TL			Effect Size
	Mean	SD	Range	Mean	SD	Range	d
NDW*	29.8	28.1	(0, 91)	66.5	50.0	(3, 166)	−0.90
TNW*	81.2	80.7	(0, 257)	291.6	272.9	(3, 1167)	−1.05
MLUm	1.58	0.73	(1, 3.84)	2.02	1.12	(1, 4.57)	−0.46
% Intelligibility	55.9	22.5	(11,100)	63.6	15.7	(29, 100)	−0.40
Caregiver–Child Play Session							
NDW*	20.9	17.5	(0, 57)	33.3	20.7	(5, 71)	−0.64
TNW*	39.1	32.2	(0, 92)	81.4	58.6	(8, 185)	−0.89
MLUm	1.43	0.52	(1, 2.8)	1.83	0.83	(1, 3.71)	−0.57
% Intelligibility	45.2	22.8	(0, 78)	54.2	14.7	(28, 85)	−0.47

*$p < .05$

Differences in Spoken Language
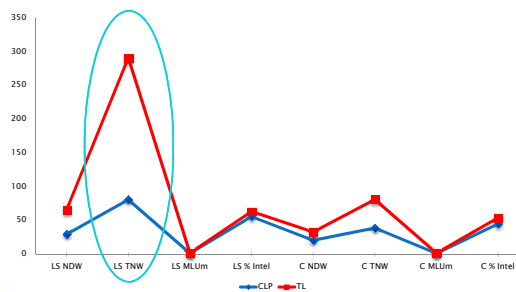
Differences in Use of Spoken Language

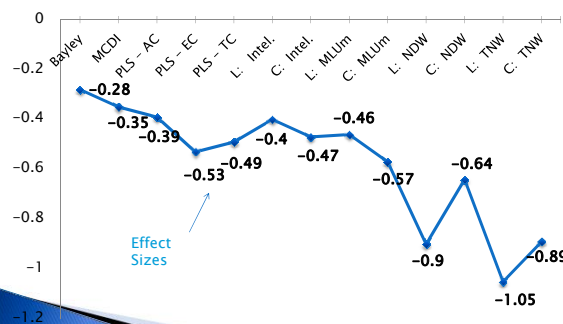| Language Sample	Play with Caregiver

- Children with CLP used fewer words
  - NDW ($F(1, 46) = 9.788$, $p = .003$)
  - TNW ($F(1, 46) = 13.122$, $p = .001$)
	- Children with CLP used fewer words
	  - NDW ($F(1, 45) = 4.87$, $p = .033$)
	  - TNW ($F(1, 45) = 9.27$, $p = .004$)

Another Look at the Differences:
Children with CLP relative to Typical Children

Functional Language Skills

‣ Significant differences observed in spoken language of children with and without CLP suggest a possible functional language deficit for young children with CLP
  ◦ Lower rate of talking
  ◦ Less diversity in spoken vocabulary

‣ Communication partner matters
  ◦ Caregiver vs examiner
  ◦ Adult vs peer (expected from parent report)

Challenges in Use of Functional Language

‣ Lower rates of talking or use of fewer words may:
  ◦ limit opportunities for interactions with peers and adults
  ◦ influence type of responses and linguistic input provided to young children with CLP by parents and/or teachers

‣ → Implications for assessment
‣ → Implications for intervention

Implications for Assessment

‣ Measurement context
‣ Measurement type
‣ Timing of measures
  ◦ Early measures may present a more optimistic picture of language development in the current sample

Implications for Early Intervention

‣ Address gap between language competence and language performance
  ◦ Increase language productivity
  ◦ Increase complexity of spoken language
  ◦ Increase spoken communication with less familiar conversational partners

‣ Cross setting language support

‣ Speech and language intervention in multiple contexts

Effects of EMT+PE on the Language Skills of Young Children with Cleft Palate

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Enhanced Milieu Teaching PLUS Phonological Emphasis (EMT+PE)

‣ Hybrid Model that teaches speech and language in a naturalistic, play based interaction
‣ **Uses the strategies of EMT (Kaiser, 1993)**
  ◦ Environmental Arrangement
  ◦ Responsive Interaction to increase engagement and talk : engage, respond, mirror + map actions
  ◦ Modeling and Semantic Expansions for words + phrases at target level
  ◦ Milieu Teaching Prompts: Time Delay, Mand, Model embedded in the system of least prompts hierarchy
‣ **Adds phonological recasting and expansions of target sounds in words (Scherer & Kaiser, 2010)**
      Model
      Repeat
      Recast for correctness
   Expand (repeat sound in whole word or phrase)

EMT + PE is an individualized, play-based intervention

- Target words selected based on both vocabulary and speech goals
- General goals:
  - Increase rate of talking, using words and sounds
  - Increase diversity of forms (more words, more sounds)
  - Increase complexity (combine words, extend sound combinations)
  - Teach for ease of generalization to conversation
- Specific goals:
  - Speech sounds not mastered: developmental sequence for complexity, position in word

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Model of Early Speech/Language Development in Children with Clefts

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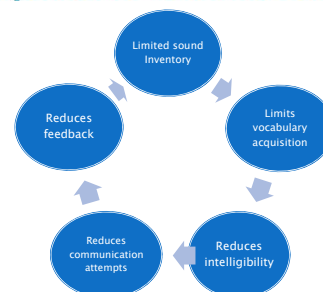
Model of Early Speech/Language Development in Children with Clefts

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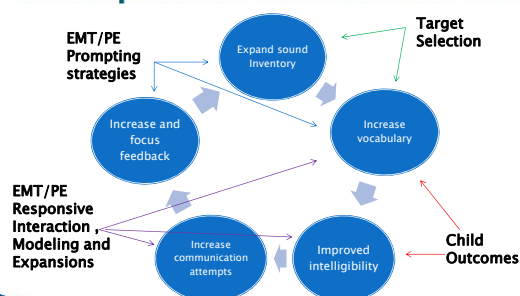
Video example of EMT +PE

Note: Consent obtained for photography and video

Study Purpose

- Investigate effects of EMT+PE intervention on expressive and receptive language skills of toddlers with CLP
- Stratified, randomized group comparison design (pilot study)
- Research Question
  - At the end of intervention, do toddlers who receive EMT+PE intervention have better language skills than children who do not receive the EMT+PE intervention?

Inclusion Criteria

- Children with repaired CLP
  - First palate repair initiated by 12 months
  - Any type of CLP accepted
- No evidence of genetic condition
- Between ages of 15 and 30 months
- Minimum of five reported words (MCDI)
- English spoken as first language in home
- Parent consented to participation

Participants

- 19 children with CLP
  - Subset of children with CLP described in first presentation
- Participants were stratified by age and gender and randomized to intervention (EMT+PE) and business-as-usual (BAU) groups
  - 8 children in EMT+PE intervention group
  - 11 children in BAU group

Description of Groups

	EMT/PE (N= 8)	BAU (N= 11)
Gender		
Male	5	8
Female	3	3
Cleft Type		
Cleft Palate	1	2
Unilateral CLP	5	6
Bilateral CLP	2	3
Race/Eth nicity	8 Caucasian	10 Caucasian 1 African American

Pre-Test Child Participant Characteristics

	Intervention Mean	Control Mean
Child's age (months)	24.3 (7.1)	26.6 (7.2)
Bailey Cognitive subscale score	96.9 (7.5)	103.2 (11.5)
Age of Palate Repair	11.5 (1.9)	11.1 (1.4)
Total # of Words on MCDI	182.0 (206.1)	303.9 (209.1)

EMT+PE Intervention Condition

- Sessions conducted in clinic room with child preferred toys and activities
- Individualized speech and language targets
- 48 Intervention sessions
- 30 minutes in length
- Session conducted by an speech language pathologist trained in EMT+PE
  - Fidelity assessed in 20% of sessions for all children
  - Criterion levels of each component of EMT+ PE including modeling of child speech and language targets

Criterion and Fidelity Levels for Intervention Components

Fidelity Measure	% Criterion	% Intervention Mean (SD)
Matched turns	>75	98 (2.8)
Talk at child's level	>50	83(12.5)
Recasted incorrect child utterances	>40	76(16.7)
Expanded child utterances	>40	55(16.0)
Time delay strategies	>80	98(8.0)
Prompting strategies	>80	98(12.4)
Words containing speech targets	>25%	34(17.3)

Language Outcomes

- **PLS-4**
  - Expressive Communication raw score
  - Auditory Comprehension raw score
- **Language Sample**
  - NDW
  - MLUm
- **Combined Language Sample + Caregiver-Child Interaction Session**
  - NDW per minute
  - MLUm
- **LENA**
  - Number of vocalizations per minute
- **MCDI (parent report)**
  - total number of words

- *Data analyzed using ordinary least squares (OLS) regression models*
  - Covariates
    - Pre-test scores
    - Child age at pre-test
    - Experimental group

Pre-Test Language

- On average, children in BAU group performed better than children in EMT+PE group across all measures at start of study
  - No statistically significant differences between groups
    - Expressive language skills
    - Receptive language skills
    - NDW per minute
    - MLUm
    - Number of LENA vocalizations per min
    - Speech intelligibility
  - Significant difference on total language skills
    - PLS-4, $F(1, 17) = 5.89$, $p = 0.027$

Intervention and Control at Pre-test

|	Intervention	Control
PLS-IV AC RS	25.1 (5.4)	31.2 (9.8)
PLS-IV AC SS**	87.4 (6.6)	104.9 (17.2)
PLS-IV EC RS	28.4 (6.7)	33.6 (8.1)
PLS-IV EC SS	94.1 (12.3)	106.4 (15.0)
PLS-IV Total Language SS*	90.0 (10.0)*	106.2 (16.8)*
MCDI total words	182.0 (206.1)	303.9 (290.1)
NDW	53.5 (51.6)	81.1 (58.8)
MLUm*	1.2 (0.4)	1.6 (0.7)

Language Outcomes

- When controlling for language skills and age at the start of the study,
  - Significant differences in receptive language skills (PLS-AC) at end of intervention were observed
  - Differences in expressive language skills between groups at the end of intervention approached significance
  - Effect sizes ranged from .04 (MLUm) to .43 (PLS-AC)
    - All ES favored intervention group
    - Effects sizes for expressive and receptive language, NDW, and MCDI were >.30

Language Outcomes

Outcome Measure	β	t	p	95% CI	d
PLS-EC RS	3.46	2.02	0.63	[-0.22, 7.14]	0.37
PLS-AC RS*	**3.91**	**2.59**	**.021**	**[0.69, 7.13]**	**0.43**
MCDI: Total Words	95.8	1.17	.264	[-81.5, 273.1]	0.40
Aggregated NDW/min	0.64	0.88	.395	[0-.92, 2.21]	0.32
Aggregated MLUm	0.05	0.12	.906	[-0.76, 0.85]	0.04
Language Sample NDW	23.06	1.31	.210	[-14.49, 60.61]	0.45
Language Sample MLUm	0.12	0.26	.802	[-0.91, 1.15]	0.05

$*p < .05$

Total Words: Pre to Post
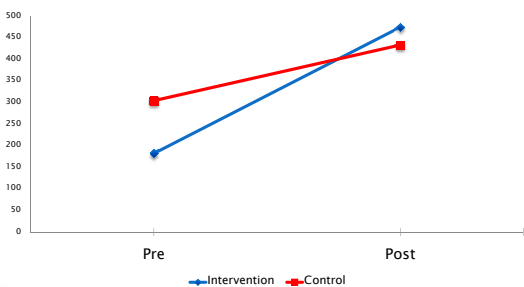
Clinical Impact of Language Findings

- On average, children who received EMT+PE intervention answered about *4 more items* correct on the AC and on the EC scales of the PLS-4 than children who did not receive intervention
- Effect sizes and clinical effects for all language measures were positive
  - 40 more words on MCDI
  - 20 more unique words (NDW) in LS
- Effect sizes for MLU were near zero

Discussion

- Study provides preliminary evidence of the effectiveness of early naturalistic intervention promoting improved language outcomes
- Study needs to be replicated with larger sample
- Child engagement and overall responsiveness was high within sessions, suggesting this is a developmentally appropriate treatment
- Parent satisfaction with the treatment was very high, indicating acceptability of the treatment

Limitations

- N for the study is extremely small
  ◦ Randomization did fully distribute differences
- Effect size differences but only one statistically significant outcome
  ◦ Sample size
  ◦ Variability across children
- 6 month follow up data have not yet been analyzed to examine maintenance of effects
- Measurement context is an issue
  ◦ Limitations of LENA to discriminate words
  ◦ Low rates of talking in LS

Effects of EMT +PE on the Speech Development Skills of Young Children with CLP

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ASU

Stages of Phonological Acquisition

1. Prelinguistic Stage (birth to 1 year)
2. First Words Stage (1 year to 18 months)
   - Early words learned as whole units (not sequence of segments)
   - Consonant production variable
   - Active selection and avoidance strategies used
3. Phonemic Development Stage (18 months to 4 years)
4. Stabilization of the Phonological System Stage (4 to 8 years)

Stoel-Gammon & Dunn, 1985

Purpose

- To assess the efficacy of an early intervention "Enhanced Milieu Teaching with Phonological Emphasis (EMT + PE)" on the speech and language development of children with CLP under 3 years of age.
  ◦ 19 children were randomly assigned to the EMT+PE intervention or a "business as usual" (BAU) control

Today's presentation

- Speech and language measures pre and post intervention
  ◦ 19 children with CLP who have completed the intervention
    · 8 children in the EMT+ PE intervention
    · 11 children in the BAU
- Compare to normative speech measures
  ◦ 40 noncleft children at 18, 24, 30 and 36 months

Speech Measures

- Pre-Post Assessment
  - Profiles of Early Expressive Phonological Skills (PEEPS)
  - Language sample
    - Clinician-child (Play)
    - Parent-child (Play, book, snack)

Profiles of Early Expressive Phonological Skills
(PEEPS: Williams & Stoel-Gammon)

- Assesses developmentally appropriate sound production in single words
  - consonant inventory
  - place/manner of articulation
  - syllable structure
  - accuracy
  - error patterns
- 18-36 months of age
- Elicited with objects

Construction of PEEPS

- 40 words
- The words were selected based on
  - age of acquisition (AOA) based on vocabulary words from the MacArthur Communicative Development Inventories
  - phonetic characteristics to elicit target English consonants across all place, voice, and manner categories of production, as well as in different syllable structures and word position.

PEEPS Pre-Post Treatment Data

- Consonant Inventory
  - Initial
  - Medial/Final
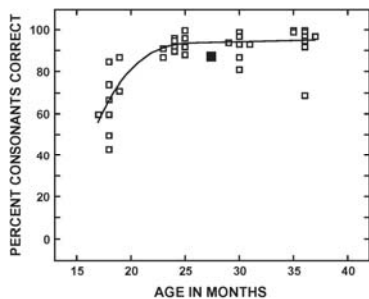- Percent Consonants Correct
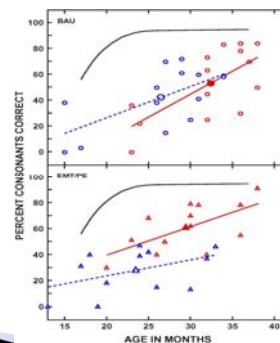- Compensatory substitutions

Speech Outcomes

Pre-Post Speech Measures

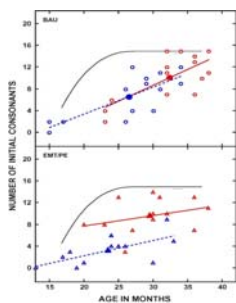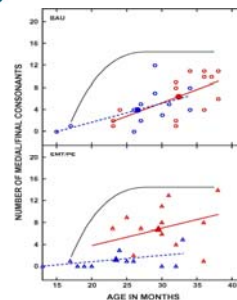| Outcome Measure	t	p	Effect Size
Percent Intelligibility	2.49	0.037	0.62
Total PCC	2.53	0.025	0.47

PCC Noncleft children
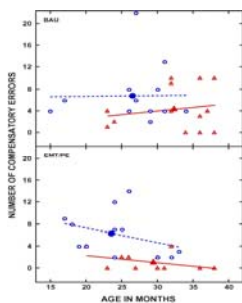

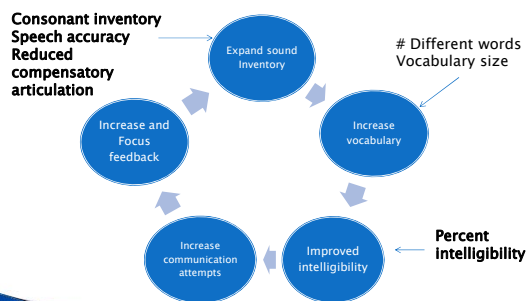## Percent Consonants Correct

Number of Initial Consonant Tokens

Number of Medial/Final Consonant Tokens

Compensatory Errors

Results informing the Model of Early Speech/Language Development

Consonant inventory
Speech accuracy
Reduced compensatory articulation

Different words
Vocabulary size

Percent intelligibility

Summary and Implications for Practice

Clinical Implications

- Assessment strategies must be developmentally appropriate
- Significant changes were found in both speech outcomes and vocabulary
- Use naturalistic techniques to facilitate communication attempts which provide the opportunity for expansions, recasts
- Must select targets that integrate vocabulary and speech goals simultaneously
- Train parents to implement naturalistic techniques

Further Considerations

- Age and number of words produced at entry to treatment
  ◦ 18 months
  ◦ 10–20 words
- Need for longitudinal research
  ◦ The trajectory of the control group suggests that children fall further behind over time, without intervention
  ◦ Longitudinal descriptions of speech, language and transition to reading are needed
  ◦ Impact on peer relationships and academic participation
- Relative need for intervention with this population
  ◦ With the 1.3 SD of normal, but consistently low

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