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A Preliminary Examination of Expressive Writing in Boys with Isolated Orofacial Clefts

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Introduction

Orofacial clefts are the most common form of craniofacial disorders in the United States, occurring in 10.25 per 10,000 live births (1). Most clefts occur in the absence of identifiable genetic abnormalities (2) and are commonly referred to as *isolated cleft of the lip and/or palate* (iCL/P). However, the significantly high incidence of language and reading concerns among children with iCL/P (3) suggests that the cleft is not truly occurring in isolation.

Given this increased risk, language and reading skills have been extensively studied (4). Despite the plethora of work looking reading and the strong correlation of these skills to writing, very few studies have explored expressive writing skills in iCL/P. Only three published research studies have specifically focused on the written language skills of patients with cleft. Ebert, McWilliams, & Woolf (5) found that children with isolated clefts of the palate only (iCPO) generally had intact, age-appropriate writing skills. However, Kommers & Sullivan (6) found lower-than-expected written language skills in children with iCPO, including fewer total words, fewer words per sentence, and poorer syntax development. Unfortunately, these studies are significantly dated (1970s), did not assess technical aspects of writing (e.g., grammar, structure), and contradict each other. A recent study by Alighieri and colleagues (7) evaluated the reading and writing skills of 12 Dutch-speaking children with iCL/P compared to matched controls. No group differences in reading or writing were found, but writing skills were correlated to working memory.

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The lack of research in this critical area limits the extent to which clinicians can provide appropriate screening, assessment, and interventions. The purpose of the current study was to obtain preliminary data on writing skills and related cognitive processes among children with isolated oral clefts compared to a sample of unaffected participants with either average or impaired reading. Results will provide preliminary guidance on future work to improve understanding of the written expression skills of individuals with iCL/P.

Methods

Participants

The 23 male participants (aged 8 to 11) in this study were drawn from a parent study evaluating reading outcomes of boys with iCL/P (8). Participants were recruited into three groups: unaffected boys with average reading (uAR, $n = 8$), unaffected boys with impaired reading (uIR, $n = 8$), and boys with iCL/P irrespective of reading ability ($n = 7$). Unaffected impaired readers (with either a confirmed diagnosis of dyslexia or an Individualized Education Plan with a reading goal) were recruited through local dyslexia support groups. Unaffected average readers (with no history of learning or attention disorders or academic support) were recruited through local advertisements. Recruitment for participants with iCL/P occurred through the Cleft Clinic at the University of Iowa. There were 3 participants with cleft lip and palate (2 bilateral and 1 unilateral left), 3 participants with cleft lip only (2 unilateral left and 1 unilateral right), and 1 participant with cleft palate only (soft palate).

The mean age across the three groups (uAR = 9.56[0.68]; uIR = 10.44[1.01]; iCL/P = 10.01[1.20]) was not significantly different ($F(2, 20) = 1.607, p = .225$). Parent-reported socioeconomic status (SES) was based on a modified 5-point Hollingshead rating scale (9), with lower scores indicating higher socioeconomic status. There were no significant group differences on SES (uAR = 2.00[0.00]; uIR = 2.25[0.46]; iCL/P = 2.29[0.49]; $F(2, 20) = 1.283, p = .299$). Most participants were White (83%) and of non-Hispanic/Latinx ethnicity (83%).

Protocol

All procedures were approved by the Institutional Review Board at the University of Iowa. Parents provided written consent and participants provided written and/or verbal assent. Monetary compensation and reimbursement for travel expenses were provided. The 3 hour protocol was completed in a single visit.

Reading—Selected subtests from the *Woodcock Reading Mastery Test, 3rd Edition* (WRMT-III; 10) were administered. Word Identification (WI) measured accuracy of single word recognition and Word Attack (WA) measured accuracy in decoding isolated nonsense words. The Oral Reading Fluency (ORF) subtest measured accuracy and speed of reading.

Intelligence & Cognitive Processes

Intelligence. Select subtests from the *Wechsler Intelligence Scale for Children, 5th Edition* (WISC-V; 11) were administered to obtain an overall General Ability Index (GAI; composite of verbal, visual, reasoning, working memory, and processing speed measures).

Given the specific language interest, the Verbal Comprehension Index (VCI; composite of Vocabulary and Similarities) was also obtained.

Auditory Memory (AM): Rote auditory memory was assessed with the Digits Forward subtest from the WISC-V and the Nonword Repetition subtest from the *Comprehensive Test of Phonological Processing* (CTOPP; 12). Both tasks require rote repetition of verbal stimuli. A mean of these subtests was calculated for a composite score.

Phonological Awareness (PA): Sound deletion (Elision) and sound blending (Blending Words) tasks were taken from the CTOPP (12) to measure phonological awareness. A mean of these subtests was calculated for a composite score.

Orthographic Competence (OC): The Homophone Choice subtest from the *Test of Orthographic Competence* (TOC; 13) was used as a single measure of orthographic competence.

Written Expression—The Essay Composition subtest from the *Wechsler Individual Achievement Test, 3rd Edition* (WIAT-III; 14) was administered to assess participants' written expression skills. The composite Total Score (TS) includes a measure of written verbosity (Word Count [WC]) and a measure of the quality, clarity, organization, and elaboration of the writing sample (Theme Development and Text Organization [TD]). A supplemental measure of appropriate syntax, spelling, capitalization, and punctuation (Grammar and Mechanics [GM]) was also obtained.

Analyses

An *a priori* analysis of reading was conducted to confirm group membership; it was anticipated that the uAR group would have the highest scores across all reading measures, with scores for the uIR group being the lowest and the iCL/P group in the middle. Next, analysis of variance (ANOVA) was used to compare the three groups (i.e., uAR, uIR, and iCL/P) on scores for cognitive processes (GAI, VCI, AM, PA, OC) and written language achievement (TC, WC, TD, GM). Brown-Forsythe and Games-Howell statistics were used if homogeneity of variance was violated. Finally, separate Pearson Correlations were run between measures of written expression (WC, TD, GM) and cognitive processes (GAI, VCI, AM, PA, OC) for the 3 participant groups (uAR, uIR, and iCL/P).

Results

Reading Status

The *a priori* analysis confirmed that the uAR group demonstrated the highest reading scores, with mean standard scores for WI, WR, and ORF all within the upper half of the average range. No uAR participants had reading scores at or below the 25th percentile. The next highest performing group was the iCL/P group; with performance in the lower half of the average range. Three participants with iCL/P had at least one reading measure at or below the 25th Percentile. Lastly, the most impaired performance was displayed by the uIR group;

with below-to-low average performance (See Table 1). All uIR participants had at least one reading measure at or below the 25th percentile.

Cognitive Processes and Expressive Writing

Among the ANOVAs run on intelligence and cognitive process, significant group differences were only found for OC ($F(2, 20) = 9.123, p = .002$). Participants in the uIR group had significantly lower scores than the uAR group (mean difference = $-4.75, p = .001$). All comparisons for written expression reached significance, with participants in the uIR group performing lower than the uAR group on all measures; differences from the iCL/P group only reached significance for GM (See Table 1).

For participants in the uAR group, the only correlation that reached significance was between GAI and GM ($r = .741, p = .036$). For participants with uIR, GM was also significantly correlated to GAI ($r = .725, p = .042$), as well as PA ($r = .763, p = .028$). For participants with iCL/P, GAI was significantly correlated to both WC ($r = .909, p = .005$) and GM ($r = .878, p = .009$). VCI was also significantly correlated to GM ($r = .783, p = .037$; See Table 2).

Discussion

Although exploratory, this study is one of the few, and the first in over 40 years, to report English written language performance in relation to cognitive processes for individuals with iCL/P. Participants with iCL/P had both cognitive processes and expressive writing skills within the average range. This is in line with findings by Ebert (5) and Alighieri (7), but contrasts those of Kommers & Sullivan (6). Results must be interpreted cautiously given the small sample size and inclusion of only one participant with iCPO. Previous research has suggested a cleft type effect, where boys with iCPO have higher risk for language disorders while boys with iCLO perform at or above the average range (8, 15–17).

The design of this study and results offer useful information for future research. The inclusion of two contrast groups provided the opportunity to evaluate if patterns of deficits among students with iCL/P mirrored that of those who were unaffected. In the current study, global ability was a stronger predictor of writing scores for participants with iCL/P and specific cognitive processes (i.e., phonological awareness) was a stronger predictor for uIR. The correlation to working memory found by Alighieri (7) was not replicated. Additionally, while participants in the uIR group demonstrated their lowest performance on Grammar/Mechanics, all measures of written expression were relatively equal among participants with iCL/P – consistent with the pattern in the uAR group.

This lends support to the hypothesis that learning disorders among participants with iCL/P may be driven by a global language deficit rather than specific cognitive processes, as seen in dyslexia. Previous work by Richman has found global language deficits to be stronger predictors of reading outcome, particularly for those with iCPO (18–20). However, some studies have identified correlations between more specific cognitive processes such as auditory memory (7, 21), phonological awareness, automaticity, and attention (8) as well as speech (22). Future research must include assessment of global and specific cognitive

processes in addition to achievement to best ascertain what may be driving any deficits and guide appropriate interventions. This work will also require larger samples to permit analysis across different cleft types.

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Impact Statement:

- Research in children with isolated cleft of the lip and/or palate (iCL/P) has demonstrated higher rates of language and reading disorders
- No work has assessed written expression in children with iCL/P in over 40 years
- This study is the first to evaluate elements of written expression and associated cognitive processes among children with iCL/P in comparison to unaffected children with either average or impaired reading skills
- Measures of writing were within the average range for children with iCL/P and demonstrated correlation to global cognitive reasoning rather than to specific cognitive processes as found in unaffected children with impaired reading

Table 1.
Cognitive Processes, Reading, and Writing Standard Scores

	uAR	uIR	iCL/P	F	p
Reading					
Word Identification	107.87 (7.97)	^a 82.25 (15.41)	96.86 (14.69)	7.768	.003
Word Attack	110.13 (10.56)	^b 83.38 (13.94)	92.43 (17.70)	7.345	.004
Oral Reading Fluency	109.38 (16.45)	^c 79.75 (7.46)	95.14 (15.66)	9.355	.001
Cognitive Processes					
General Ability	111.13 (3.72)	109.50 (12.17)	103.43 (9.95)	* 1.384	.282
Verbal Comprehension	108.13 (4.94)	106.71 (14.84)	97.14 (11.20)	* 2.054	.169
Auditory Memory	10.13 (1.43)	7.81 (1.49)	9.43 (2.44)	3.420	.053
Phonological Awareness	11.44 (1.72)	9.38 (1.03)	10.93 (2.47)	2.831	.083
Orthographic Competence	11.00 (1.85)	^d 6.25 (2.12)	9.14 (2.73)	9.123	.002
Written Expression					
Total Score	112.25 (4.95)	^e 92.50 (7.80)	110.71 (19.30)	* 4.998	.036
Word Count	111.13 (7.90)	^f 93.00 (6.61)	101.86 (18.58)	* 4.249	.048
Theme/Development	111.25 (5.04)	^g 91.63 (11.45)	99.43 (20.15)	4.419	.026
Grammar/Mechanics	115.00 (6.85)	^h 71.25 (9.62)	99.86 (18.28)	* 24.362	< .001

Note: uAR = Unaffected and Average Reading; uIR = Unaffected and Impaired Reading; iCL/P = Isolated Cleft Lip and/or Palate.

* Utilized Brown-Forsythe and Games-Howell due to inequality of variance.

^a uIR < uAR (mean difference = -25.625, p = .002);

^b uIR < uAR (mean difference = -26.750, p = .004);

^c uIR < uAR (mean difference = -29.63, p = .001);

^d uIR < uAR (mean difference = -4.750, p = .001);

^e uIR < uAR (mean difference = -19.750, p < .001);

^f uIR < uAR (mean difference = -18.125, p = .001);

^g uIR < uAR (mean difference = -19.625, p = .024);

^h uIR < uAR and iCL/P (mean difference = -43.750 & -28.607, p < .001 and p = .012, respectively).

Table 2.

Pearson Correlations (r) Between Cognitive Processes and Written Expression

	uAR			uIR			iCL/P		
	WC	TD	GM	WC	TD	GM	WC	TD	GM
General Ability	.554	.364	.741	-.174	.489	.725	.909	.600	.878
Verbal Comprehension	.076	.538	.084	-.409	.089	.666	.629	.373	.783
Auditory Memory	-.058	-.272	.087	.48	.432	.114	-.223	-.001	.187
Phonological Awareness	-.083	.216	.091	-.484	-.278	.763	.477	.325	.615
Orthographic Competence	-.615	.444	.023	-.387	-.401	.305	.345	-.171	.441

Note: uAR = Unaffected and Average Reading; uIR = Unaffected and Impaired Reading; iCL/P = Isolated Cleft Lip and/or Palate; WC = Word Count; TD = Theme/Development; GM = Grammar/Mechanics. **Bold** r-values indicate significance at $p < .05$.