www.nature.com/pr

Pediatric RESEARCH

SPECIAL ARTICLE

A preliminary examination of expressive writing in boys with isolated orofacial clefts

Jon W. Goodwin[1] and Amy L. Conrad[2]

BACKGROUND: Children with isolated cleft of the lip and/or palate (iCL/P) are at a higher risk for language and reading issues. The current pilot study evaluated concurrent writing skills of children with iCL/P compared to unaffected participants with average (uAR) and impaired (uIR) reading. It was hypothesized that children with iCL/P would perform lower than age-expectations.
METHODS: Twenty-three males, aged 8–11 years old, were recruited through clinics, local advertisements, and state dyslexia groups (iCL/P = 7, uAR = 8, uIR = 8). Group differences on measures of cognitive processes and writing were evaluated using ANCOVA. Relationships between these measures were compared for each group through Pearson correlations.
RESULTS: Participants with iCL/P performed within the average range across all measures; group differences were only found for the uIR group. For those with iCL/P, writing was correlated to global cognitive skills rather than more specific skills.
CONCLUSIONS: While this small sample of children with iCL/P demonstrated average writing skills, patterns suggest performance is related to global cognitive reasoning rather than specific cognitive processes as found in unaffected children with impaired reading. Further research is needed to better understand writing in iCL/P and the relationship to reading and cognitive processes.

Pediatric Research (2022) 91:1370–1373; https://doi.org/10.1038/s41390-021-01619-y

IMPACT:

- 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.

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 at 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, and Woolf[5] found that children with isolated clefts of the palate only (iCPO) generally had intact, age-appropriate writing skills. However, Kommers and 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.

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.

[1]Department of Counseling, Clinical, and School Psychology, University of California Santa Barbara, Santa Barbara, CA, USA and [2]The Stead Family Department of Pediatrics, University of Iowa Roy J and Lucille A Carver College of Medicine, Iowa City, IA, USA
Correspondence: Jon W. Goodwin (jongoodwin@ucsb.edu)

Received: 12 February 2021 Revised: 27 April 2021 Accepted: 26 May 2021
Published online: 11 June 2021

METHODS

Participants

The 23 male participants (aged 8–11 years) 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 three participants with cleft lip and palate (two bilateral and one unilateral left), three participants with cleft lip only (two unilateral left and one unilateral right), and one 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 ($F^{2,20} = 1.607$, $p = 0.225$). Parent-reported socioeconomic status (SES) was based on a modified five-point Hollingshead rating scale,[9] with lower scores indicating higher SES. There were no significant group differences on SES (uAR = 2.00 [0.00]; uIR = 2.25 [0.46]; iCL/P = 2.29 [0.49]; $F^{2,20} = 1.283$, $p = 0.299$). 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-h protocol was completed in a single visit.

Reading. Selected subtests from the Woodcock Reading Mastery Test, Third Edition[10] were administered. Word Identification (WI) measured accuracy of single word recognition, and Word Attack measured accuracy in decoding isolated nonsense words. The Oral Reading Fluency (ORF) subtest measured accuracy and speed of reading.

Intelligence and cognitive processes

Intelligence: Select subtests from the Wechsler Intelligence Scale for Children, Fifth 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 AM 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 PA. 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 OC.

Written expression. The Essay Composition subtest from the Wechsler Individual Achievement Test, Third Edition[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 three 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 ($F^{2,20} = 9.123$, $p = 0.002$). Participants in the uIR group had significantly lower scores than the uAR group (mean difference = −4.75, $p = 0.001$). 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 ($r = 0.741$, $p = 0.036$). For participants with uIR, GM was also significantly correlated to GAI ($r = 0.725$, $p = 0.042$), as well as PA ($r = 0.763$, $p = 0.028$). For participants with iCL/P, GAI was significantly correlated to both WC ($r = 0.909$, $p = 0.005$) and GM ($r = 0.878$, $p = 0.009$). VCI was also significantly correlated to GM ($r = 0.783$, $p = 0.037$; 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 et al.[5] and Alighieri et al.[7] but contrasts those of Kommers and 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]

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**	**0.003**
Word Attack	110.13 (10.56)	[b]83.38 (13.94)	92.43 (17.70)	**7.345**	**0.004**
Oral Reading Fluency	109.38 (16.45)	[c]79.75 (7.46)	95.14 (15.66)	**9.355**	**0.001**
Cognitive Processes					
General Ability	111.13 (3.72)	109.50 (12.17)	103.43 (9.95)	[i]1.384	0.282
Verbal Comprehension	108.13 (4.94)	106.71 (14.84)	97.14 (11.20)	[i]2.054	0.169
Auditory Memory	10.13 (1.43)	7.81 (1.49)	9.43 (2.44)	3.420	0.053
Phonological Awareness	11.44 (1.72)	9.38 (1.03)	10.93 (2.47)	2.831	0.083
Orthographic Competence	11.00 (1.85)	[d]6.25 (2.12)	9.14 (2.73)	**9.123**	**0.002**
Written Expression					
Total Score	112.25 (4.95)	[e]92.50 (7.80)	110.71 (19.30)	[i]**4.998**	**0.036**
Word Count	111.13 (7.90)	[f]93.00 (6.61)	101.86 (18.58)	[i]**4.249**	**0.048**
Theme/Development	111.25 (5.04)	[g]91.63 (11.45)	99.43 (20.15)	**4.419**	**0.026**
Grammar/Mechanics	115.00 (6.85)	[h]71.25 (9.62)	99.86 (18.28)	[i]**24.362**	**<0.001**

uAR unaffected and average reading, *uIR* unaffected and impaired reading, *iCL/P* isolated cleft lip and/or palate.
Statistically significant $p < 0.05$ values are in bold.
[a]uIR < uAR (mean difference = −25.625, $p = 0.002$).
[b]uIR < uAR (mean difference = −26.750, $p = 0.004$).
[c]uIR < uAR (mean difference = −29.63, $p = 0.001$).
[d]uIR < uAR (mean difference = −4.750, $p = 0.001$).
[e]uIR < uAR (mean difference = −19.750, $p < 0.001$).
[f]uIR < uAR (mean difference = −18.125, $p = 0.001$).
[g]uIR < uAR (mean difference = −19.625, $p = 0.024$).
[h]uIR < uAR and iCL/P (mean difference = −43.750 and −28.607, $p < 0.001$ and $p = 0.012$, respectively).
[i]Utilized Brown–Forsythe and Games–Howell due to inequality of variance.

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	0.554	0.364	**0.741**	−0.174	0.489	**0.725**	**0.909**	0.600	**0.878**
Verbal Comprehension	0.076	0.538	0.084	−0.409	0.089	0.666	0.629	0.373	**0.783**
Auditory Memory	−0.058	−0.272	0.087	0.48	0.432	0.114	−0.223	−0.001	0.187
Phonological Awareness	−0.083	0.216	0.091	−0.484	−0.278	**0.763**	0.477	0.325	0.615
Orthographic Competence	−0.615	0.444	0.023	−0.387	-0.401	0.305	0.345	−0.171	0.441

Bold *r*-values indicate significance at $p < 0.05$.
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.

The design of this study and results offer useful information for future research. The inclusion of two contrast groups provided the opportunity to evaluate whether 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., PA) was a stronger predictor for uIR. The correlation to working memory found by Alighieri et al.[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 AM,[7,21] PA, 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.

ACKNOWLEDGEMENTS

The authors would like to express gratitude to Emily Kuhlmann, BA for her assistance with data collection. Additional thanks are owed to the chapter leaders at Decoding Dyslexia and the International Dyslexia Association for facilitating successful recruitment. Lastly, thank you to the families who participated in this study. This study was supported by grants from the National Institute of Dental and Craniofacial

Research (DE024511; to A.L.C.) and the National Center for Advancing Translational Sciences (UL1TR002537).

AUTHOR CONTRIBUTIONS

J.W.G.: contributed to design of the work, analysis and interpretation of data, and revision and final approval of the manuscript. A.L.C.: contributed to design of the work; acquisition, analysis, and interpretation of data; and revision and final approval of the manuscript.

ADDITIONAL INFORMATION

Competing interests: The authors declare no competing interests.

Patient consent: Parents/guardians provided written consent and children provided verbal and/or written assent to participate.

Publisher's note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

REFERENCES

1. Mai, C. T. et al. National population-based estimates for major birth defects, 2010-2014. *Birth Defects Res.* **111**, 1420–1435 (2019).
2. Jones, M. C. & Jones, K. L. in *Comprehensive Cleft Care* (eds Losee, J. & Kirshner, R.) 107–127 (McGraw-Hill, 2009).
3. Richman, L. C., McCoy, T. E., Conrad, A. L. & Nopoulos, P. C. Neuropsychological, behavioral, and academic sequelae of cleft: early developmental, school age, and adolescent/young adult outcomes. *Cleft Palate Craniofac. J.* **49**, 387–396 (2012).
4. Conrad, A. L., Richman, L. & Nopoulos, P. Reading achievement in boys with non-syndromic cleft palate only: relationship to neuropsychological skill and neuro-circuitry. *Dev. Neuropsychol.* **40**, 395–406 (2015).
5. Ebert, P. R., McWilliams, B. J. & Woolf, G. A comparison of the written language ability of cleft palate and normal children. *Cleft Palate J.* **11**, 17–20 (1974).
6. Kommers, M. S. & Sullivan, M. D. Written language skills of children with cleft palate. *Cleft Palate J.* **16**, 81–85 (1979).
7. Alighieri, C. et al. Technical reading and writing skills and their relationship with linguistic processes in children with a cleft (lip and) palate: a comparison with peers. *Folia Phoniatr. Logop.* https://doi.org/10.1159/000512449 (2020).
8. Conrad, A. L. Are predictors of reading impairment in isolated cleft similar to those in idiopathic dyslexia? *Ann. Dyslexia* **69**, 153–165 (2019).
9. Hollingshead, A. B. *Four Factor Index of Social Status* (Department of Sociology, Yale University, 1975).
10. Woodcock, R. W. *Woodcock Reading Mastery Tests, Third Edition Manual* (Pearson, 2011).
11. Wechsler. *Wechsler Intelligence Scale for Children* 5th edn (PsychCorp, 2014).
12. Wagner, R. K., Torgesen, J. K. & Rashotte, C. A. *Comprehensive Test of Phonological Processing* (PRO-ED, 1999).
13. Mather, N., Robers, R., Hammill, D. D. & Allen, E. A. *Test of Orthographic Competence; Examiner's Manual* (Pro-Ed, 2008).
14. Wechsler, D. *Wechsler Individual Achievement Test* 3rd edn (Psychological Corporation, 2009).
15. Collett, B. R. et al. Academic achievement in children with oral clefts versus unaffected siblings. *J. Pediatr. Psychol.* **39**, 743–751 (2014).
16. Wehby, G. L., Collett, B. R., Barron, S., Romitti, P. & Ansley, T. Children with oral clefts are at greater risk for persistent low achievement in school than classmates. *Arch. Dis. Child.* **100**, 1148–1154 (2015).
17. Conrad, A. L. et al. Retrospective evaluation of number of surgeries and parent ratings of academic and behavioral functioning among children with isolated oral clefts. *Cleft Palate Craniofac. J.* https://doi.org/10.1177/1055665620982807 (2020).
18. Richman, L. C. Cognitive patterns and learning disabilities of cleft palate children with verbal deficits. *J. Speech Hear. Res.* **23**, 447–456 (1980).
19. Richman, L. C. & Eliason, M. Type of reading disability related to cleft type and neuropsychological patterns. *Cleft Palate J.* **21**, 1–6 (1984).
20. Richman, L. C., Eliason, M. J. & Lindgren, S. D. Reading disability in children with clefts. *Cleft Palate J.* **25**, 21–25 (1988).
21. Conrad, A. L., McCoy, T. E., DeVolder, I., Richman, L. C. & Nopoulos, P. Reading in subjects with an oral cleft: speech, hearing and neuropsychological skills. *Neuropsychology* **28**, 415–422 (2014).
22. Chapman, K. L. The relationship between early reading skills and speech and language performance in young children with cleft lip and palate. *Cleft Palate Craniofac. J.* **48**, 301–311 (2011).