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SCHOOL PERFORMANCE FOR CHILDREN WITH CLEFT LIP AND PALATE: A POPULATION-BASED STUDY

Authors
Jane C Bell[1]
Camille Raynes-Greenow[1]
Robin Turner[2]
Carol Bower[3,4]
Alan Dodson[5]
Wendy Nicholls[6]
Natasha Nassar[7]

1. Sydney School of Public Health, University of Sydney, New South Wales, Australia
2. School of Public Health and Community Medicine, University of New South Wales, New South Wales, Australia
3. Western Australian Register of Developmental Anomalies, King Edward Memorial Hospital, Subiaco, Western Australia, Australia
4. Telethon Institute for Child Health Research, Centre for Child Health Research, University of Western Australia, Western Australia, Australia
5. Evaluation and Accountability, WA Education Department, East Perth, Western Australia, Australia
6. Cleft Lip and Palate and Craniomaxillofacial Unit, Princess Margaret Hospital, Subiaco, Western Australia, Australia
7. Menzies Centre for Health Policy, University of Sydney, New South Wales, Australia

Corresponding author
Jane Bell
Room 129A, Edward Ford Building A27
School of Public Health
University of Sydney
NSW 2006
Australia
Ph 61-2-9351 7789
Email: janecbell@gmail.com

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ABSTRACT

Background Educational attainment is important in shaping young people's life prospects. To investigate whether being born with orofacial cleft (OFC) affects school performance, we compared school test results between children born with and without OFC.

Methods Using record-linked datasets, we conducted a population-based cohort study of children liveborn in Western Australia 1980-2010 with a diagnosis of OFC on the Register of Developmental Anomalies, and a random sample of 6603 children born without OFC. We compared odds ratios for meeting national minimum standards in five domains (reading, numeracy, writing, spelling, grammar and punctuation), and adjusted OR (aOR) for children with cleft lip only (CLO), cleft lip and palate (CL+P) and cleft palate only (CPO) for each domain.

Results Results from two testing programs (WALNA and NAPLAN) were available for 3238 (89%) children expected to participate. Most met the national minimum standards. Compared to children without OFC, children with CPO were less likely to meet minimum standards for NAPLAN reading (aOR 0.57 [95%CI 0.34, 0.96]) grammar and punctuation (aOR 0.49 [95%CI 0.32, 0.76]), WALNA writing (aOR 0.66 [95%CI 0.47, 0.92]), and WALNA and NAPLAN numeracy (aOR 0.64 [95%CI 0.43, 0.95], and aOR 0.47 [95%CI 0.28, 0.82]) respectively. Children with CL+P had significantly lower odds for reaching the spelling standard in NAPLAN tests (aOR 0.52 [95%CI 0.29, 0.94]). Children with CLO had similar odds for reaching all minimum standards.

Conclusion Children born with OFC, particularly children with CPO, should be monitored to identify learning difficulties early, to enable intervention to maximise school attainment.

BACKGROUND

Educational attainment is important in shaping young people's life prospects.[1 2] Historically, educational outcomes for children with orofacial clefts (OFC) have been determined from small studies susceptible to selection and outcome measurement biases, with a large variety of outcome measures reported[3-5] and inconsistent findings.[3] Two population-based cohort studies reported that individuals born with isolated cleft lip and palate (CL+P) or with isolated cleft palate only (CPO) had poorer school test results.[5 6] An improved understanding of cognitive and academic outcomes associated with OFC can be used by families and school systems to monitor the progress of children with OFC and to provide additional educational support if their learning is being affected.[3] To investigate the effects of being born with OFC on school test results we conducted a population-based study including children with additional anomalies as well as children with isolated clefts.

METHODS

Data sources

In this cohort study we used linked population-based data from seven data collections in Western Australia (WA).[7] These were 1) the WA Register of Developmental Anomalies (WARDA), a population-based statutory notification system of congenital anomalies, with active surveillance from administrative data collections (such as birth and hospitalisation data) and other sources (public and private diagnostic and treatment sources). Information is collated on all reported structural and functional anomalies diagnosed in pregnancies terminated because of fetal anomaly, stillbirths and in liveborn children up to six years of age. 2) The Midwives Notification System, a legislated surveillance system covering all births in WA of more than 20 weeks gestation or 400g birthweight. 3) The Birth and 4) Death Registries which collect data on all births and deaths registered in WA. 5) The Hospital

Morbidity Data System, a census of all public and private inpatient hospital admissions. 6) The idEA database which contains population-based data on people with intellectual disability (defined as full IQ <70, or evidence of developmental delay at less than 18 years of age and accepted that the delay was present during childhood), with ascertainment from multiple sources.[8]

During the study period, standardised test records for all children attending all schools in the state were maintained by 7) the WA Department of Education. The standardised test results cover two testing programs. From 2000-07 results are from state-based assessments: Western Australian Literacy and Numeracy Assessment (WALNA) and Monitoring Standards in Education (Year 9 only). In this study, we have referred to these assessments as WALNA. In 2008 WALNA was replaced by the Australia-wide National Assessment Program – Literacy and Numeracy (NAPLAN) and results to 2011 are included. Both programs were conducted annually, examining children in school year levels 3, 5, 7 and 9 in numeracy, reading, writing and spelling domains. Grammar and punctuation was also assessed in NAPLAN tests. In 2005, the rubric for WALNA writing assessment changed. NAPLAN records also classified children as exempt from a test if they were registered with a significant and complex disability.[9]

Study population

All infants liveborn in WA between 1980 and 2010 with OFC were identified from the WARDA using the British Paediatric Association-ICD9 codes for cleft palate only (CPO) (74900-74909), cleft lip only (CLO) (74910-74919) and cleft lip and palate (CL+P) (74920-74927, 74929). If other major congenital anomalies, including chromosomal, structural or syndrome-like diagnoses (BPA-ICD9 codes 740-759) were recorded as well as OFC, these

infants were included and defined as having an additional anomaly. A comparison cohort, frequency matched 4:1 on year of birth and not diagnosed with OFC (but potentially with other major anomalies), was randomly selected from liveborn infants recorded in the Midwives Notification System. We included children who were expected to have participated in either or both testing programs and excluded children born too long ago or too recently to have school test results, children with an intellectual disability or classified as exempt, and children who died before the Year 3 test.

Study outcomes and covariates

Both programs were curriculum based, and benchmarked to a national minimum standard for each domain at each school year level.[10 11] Children not meeting these minimum standards did not have the basic elements of literacy and numeracy and were classified as needing additional support to help them achieve the skills they require to progress in schooling.[9] We used the national minimum standard for each domain as our outcome measure as this level indicates whether children require additional support.

Being born with OFC or not, having a major congenital anomaly (for those without OFC) or having an additional major anomaly (for those with OFC) were identified from the WARDA. Other datasets provided information on sex, singleton or multiple birth, birthweight (<2500g vs ≥2500g), gestational age (preterm <37 weeks vs term ≥37 weeks), family order (oldest child or not), and Indigenous status. As a measure of socioeconomic level for each child, we used quintiles of the Index of Education and Occupation based on parental residence at the time of the child's birth. This Index ranks neighbourhood areas on their educational and occupational structure from information collected at each five-yearly national Census.[12]

School test data provided information on language background (English or other language spoken at home). NAPLAN data also included information on school location (remote/very remote, regional or metropolitan), and on parental education and occupation levels recorded at the time of school enrolment. For each child we determined the highest education and occupation level from either parent. Where children had records for the same year level in consecutive calendar years (n=16 children, 58 test results), we included the most recent record. We categorised each child's age relative to their cohort at the first test result as being in the oldest, middle, or youngest third of their class. As a proxy for school absence, we calculated the number of days spent in hospital from age five years until each test date (0, 1-7, >7 days).

Statistical analyses

Participants were children who achieved a score in any domain that could be categorised as meeting the national minimum standard or not. Using Chi-square tests we compared characteristics of participants and non-participants, and then among participants, we compared characteristics of children with and without OFC, and between type of OFC. We also compared participation between WALNA and NAPLAN programs at each year level. The denominator included children expected to have a test record at each year level based on their year of birth and calendar year of test.

Using logistic regression, we calculated odds ratios (OR) and 95% confidence intervals (CI) to compare the proportion of children with and without OFC, and the proportion of children with OFC with and without an additional major anomaly, meeting the minimum standard in each domain for each school year level in each test program.

We summarised results for children with each cleft type, for each domain over all school year levels for each program using models fitted by generalised estimating equations with a logit link function, binomial distribution and robust standard errors, and accounting for within-person correlation. The *a priori* base model for each domain included test program (WALNA or NAPLAN), school year level, child's sex and Indigenous status, and quintile of parental socioeconomic level at the child's birth. To obtain the final model for each domain we added additional covariates individually to the base model using forwards selection. Significance was based on Type III likelihood ratio P-values ($p<0.05$) to test the overall effect of the variable. While the WALNA writing rubric changed in 2005, there was no association between cleft type (CLO, CL+P, CPO) and reaching the minimum standard in either of the two rubrics, so these two rubrics were included in the model with a variable adjusting for each.

As parents' education and occupation levels were only available for NAPLAN records, we conducted a sensitivity analysis for NAPLAN results for each domain, replacing the socioeconomic quintiles with these variables from NAPLAN in the final model. We also conducted a sensitivity analysis adding time spent in hospital from five years of age until the test date, for each program and domain, by school year level.

Members of CleftPALS WA, the support group for families with members affected by OFC, provided advice for this study. The study protocol was approved by the WA Department of Health Human Research Ethics Committee and the WA Aboriginal Health Ethics Committee.

RESULTS

Of the 8112 children in our cohort, we excluded 4263 born outside the testing programs, and 219 who had an intellectual disability or who died before the Year 3 test (Figure 1). The final study population comprised 3630 children expected to have test records, of whom 3238 (89.2%) participated in at least one domain; providing 33,243 test results.

Figure 1. Study cohort

Cohort		OFC	No OFC
8112	born 1980-2010	1509	6603
	excluded:		
→	*2228 born 1980-87, or born 1988 and no test record*	*401*	*1827*
→	*2035 born 2004-2010 or born 2003 and no test record*	*400*	*1635*
→	*219 with intellectual disability and, or died before Year 3 test*	*126*	*93*
3630	potential to have test records	582	3048

There were no differences in birth characteristics between participants and non-participants. Characteristics of participants with and without OFC are shown in Table 1. There were few differences between these groups but low birthweight was more common among children with OFC, and children without OFC were more likely to be the oldest child in their family. One-quarter (24.8%) of children with OFC had an additional major anomaly and 4.1% of children without OFC were born with a major anomaly. Among children with OFC, 134 were born with CLO, 145 with CL+P and 253 with CPO. Children with CLO or CL+P were more likely to be male (67.9% and 60.7% respectively), and children with CPO were more likely to be female (59.7%). Co-existing anomalies were more frequent among children with CPO (40.3%) than children with CLO (7.5%) or CL+P (13.8%). Other characteristics were evenly distributed among children with the different types of cleft. The mean age for children sitting these tests was similar for those with and without OFC (8.3, 10.3, 12.2, and 14.1 years at

school year levels 3, 5, 7 and 9 respectively). At all year levels, in both programs, scores were available for at least 80% of children.

Table 1. Characteristics of participants, with and without orofacial clefts

Characteristic	OFC N=532		No OFC N=2706		p-value
	n	(%)	n	(%)	
Males	281	(52.8)	1366	(50.5)	.32
Singleton	509	(95.7)	2628	(97.1)	.08
Preterm birth [a]	44	(8.3)	173	(6.4)	.11
Birthweight < 2500g	47	(8.8)	143	(5.3)	.001
Oldest in family[a,b]	181	(34.1)	1070	(39.5)	.02
Indigenous	45	(8.5)	181	(6.7)	.14
Language background not English[c]	92	(17.3)	422	(15.6)	.33
Age relative to class					
oldest third	193	(36.3)	864	(31.9)	
middle third	189	(35.5)	977	(36.1)	.09
youngest third	150	(28.2)	865	(32.0)	
Socioeconomic quintiles[a]					
5 highest	75	(14.3)	403	(15.0)	
4	106	(20.2)	534	(19.9)	
3	93	(17.7)	504	(18.8)	.18
2	148	(28.2)	632	(23.6)	
1 lowest	103	(19.6)	609	(22.7)	

[a] missing data: preterm birth (n=1), oldest in family (n=2), socioeconomic quintiles (n=31)
[b] at birth, oldest child alive
[c] language other than English spoken at home
OFC, orofacial cleft

The majority of students met the national minimum standard in both test programs, in all domains at each year level (Table 2). The proportion of children meeting minimum standards was slightly lower among children with OFC. In particular, the crude odds for numeracy and writing domains were significantly lower for children with OFC. Among children with OFC, the proportion of children meeting the minimum standard was similar for children with isolated OFC and for children with an additional major anomaly (Table 3), although results were imprecise due to small numbers.

Table 2. Proportion and unadjusted odds ratios[a] for reaching national minimum standards for children with OFC compared to children without OFC, by test program, domain and year level

		WALNA								NAPLAN							
		OFC			No OFC					OFC			No OFC				
Domain	Year level	N	n	%	N	n	%	OR	(95% CI)	N	n	%	N	n	%	OR	(95% CI)
Reading	3	212	196	92.5	1062	1009	95.0	0.64	(0.36, 1.13)	149	139	93.3	736	672	91.3	1.32	(0.66, 2.64)
	5	199	186	93.5	1068	996	93.3	1.03	(0.56, 1.91)	153	131	85.6	692	616	89.0	0.73	(0.44, 1.22)
	7	216	190	88.0	1146	972	84.8	1.31	(0.84, 2.03)	133	115	86.5	642	608	94.7	0.36	(0.20, 0.65)
	9	77	71	92.2	417	384	92.1	1.02	(0.41, 2.52)	115	102	88.7	568	519	91.4	0.74	(0.39, 1.42)
Numeracy	3	240	197	82.1	1215	1097	90.3	0.49	(0.34, 0.72)	149	136	91.3	736	701	95.2	0.52	(0.27, 1.01)
	5	233	194	83.3	1251	1123	89.8	0.57	(0.38, 0.84)	153	142	92.8	691	642	92.9	0.99	(0.50, 1.94)
	7	214	173	80.8	1146	919	80.2	1.04	(0.72, 1.51)	127	116	91.3	634	608	95.9	0.45	(0.22, 0.94)
	9	80	75	93.8	428	398	93.0	1.13	(0.43, 3.01)	116	104	89.7	558	528	94.6	0.49	(0.24, 0.99)
Writing[b,c,d]	3 combined	229	189	82.5	1208	1016	84.1	0.89	(0.61, 1.30)	151	142	94.0	737	701	95.1	0.81	(0.38, 1.72)
	3[b]	145	121	83.5	814	681	83.7	0.98	(0.61, 1.58)								
	3[d]	84	68	81.0	394	335	85.0	0.75	(0.41, 1.38)								
	5 combined	231	192	83.1	1236	1088	88.0	0.67	(0.46, 0.98)	155	139	89.7	693	623	89.9	0.98	(0.55, 1.73)
	5[b]	137	119	86.9	762	678	88.9	0.83	(0.48, 1.43)								
	5[d]	94	73	77.7	473	410	86.7	0.53	(0.31, 0.93)								
	7 combined	212	165	77.8	1135	951	83.8	0.68	(0.47, 0.97)	130	104	80.0	641	581	90.6	0.41	(0.25, 0.69)
	7[c]	115	82	71.3	631	506	80.2	0.61	(0.39, 0.96)								
	7[d]	97	83	85.6	504	445	88.3	0.79	(0.42, 1.47)								
	9[d]	73	70	95.9	418	382	91.4	2.20	(0.66, 7.34)	114	91	79.8	572	500	87.4	0.57	(0.34, 0.96)
Spelling[e]	3	180	141	78.3	891	725	81.4	0.83	(0.56, 1.23)	153	137	89.5	737	679	92.1	0.73	(0.41, 0.31)
	5	185	152	82.2	951	791	83.2	0.93	(0.62, 1.41)	155	135	87.1	693	630	90.9	0.68	(0.40, 1.15)
	7	175	138	78.9	964	778	80.7	0.89	(0.60, 1.33)	131	116	88.6	642	585	91.1	0.75	(0.41, 1.38)
	9									115	92	80.0	577	511	88.6	0.52	(0.31, 0.87)
Grammar & punctuation[e]	3									153	133	86.9	737	666	90.4	0.71	(0.42, 1.20)

5	155	135	87.1	693	625	90.2	0.73	(0.43, 1.25)
7	131	106	80.9	642	581	90.5	0.45	(0.27, 0.74)
9	115	94	81.7	577	501	86.8	0.68	(0.40, 1.16)

[a] Reference group is children without OFC. Children with an intellectual disability, children classified as exempt, and children who died before the year level test were excluded

[b, c, d] WALNA writing rubric changed in 2005. Results presented separately for periods 2000-04[b], 2001-04[c] and 2005-07[d]

[e] Spelling not tested at WALNA Year 9 level; Grammar & punctuation not tested in WALNA

CI, confidence interval; N, number participating; n, number reaching minimum standard; NAPLAN, National Assessment Program Literacy and Numeracy; OFC, orofacial cleft; OR, odds ratio; WALNA, Western Australian Literacy and Numeracy Program

Table 3. Proportion and unadjusted odds ratios[a] for reaching national minimum standards for children with OFC and an additional major anomaly compared to children with isolated OFC, by test program, domain and year level

		WALNA								NAPLAN							
		Isolated OFC			OFC+major anomaly					Isolated OFC			OFC+major anomaly				
Domain	Year level	N	n	%	N	n	%	OR	(95% CI)	N	n	%	N	n	%	OR	(95% CI)
Reading	3	162	149	92.0	50	47	94.0	1.37	(0.37, 5.00)	118	110	93.2	31	29	93.6	1.06	(0.21, 5.24)
	5	143	135	94.4	56	51	91.1	0.60	(0.19, 1.93)	126	111	88.1	27	20	74.1	0.39	(0.14, 1.07)
	7	155	136	87.7	61	54	88.5	1.08	(0.43, 2.71)	102	89	87.3	31	26	83.9	0.76	(0.25, 2.33)
	9	51	49	96.1	26	22	84.6	0.22	(0.04, 1.32)	85	77	90.6	30	25	83.3	0.52	(0.16, 1.73)
Numeracy	3	179	148	82.7	61	49	80.3	0.86	(0.41, 1.79)	117	109	93.2	32	27	84.4	0.40	(0.12, 1.31)
	5	164	140	85.4	69	54	78.3	0.62	(0.30, 1.27)	126	119	94.4	27	23	85.2	0.34	(0.09, 1.25)
	7	153	124	81.1	61	49	80.3	0.96	(0.45, 2.02)	98	89	90.8	29	27	93.1	1.37	(0.28, 6.70)
	9	54	51	94.4	26	24	92.3	0.71	(0.11, 4.51)	86	77	89.5	30	27	90.0	1.05	(0.27, 4.17)
Writing[b,c,d]	3 combined	170	143	84.1	59	46	78.0	0.67	(0.32, 1.40)	119	113	95.0	32	29	90.6	0.51	(0.12, 2.18)
	3[b]	104	89	85.6	41	32	78.1	0.60	(0.24, 1.50)								
	3[d]	66	54	81.8	18	14	77.8	0.78	(0.22, 2.78)								
	5 combined	163	134	82.2	68	58	85.3	1.26	(0.57, 2.74)	127	112	88.2	28	27	96.4	3.62	(0.46, 28.58)
	5[b]	96	81	84.4	41	38	92.7	2.35	(0.64, 8.59)								
	5[d]	67	53	79.1	27	20	74.1	0.76	(0.27, 2.14)								
	7 combined	153	115	75.2	59	50	84.8	1.84	(0.83, 4.08)	98	81	82.7	32	23	71.9	0.54	(0.21, 1.36)
	7[c]	80	54	67.5	35	28	80.0	1.93	(0.74, 4.99)								
	7[d]	73	61	83.6	24	22	91.7	2.16	(0.45, 10.45)								
	9[d]	48	45	93.8	25	25	100.0	-	-	85	67	78.8	29	24	82.8	1.29	(0.43, 3.86)
Spelling[e]	3	140	112	80.0	40	29	72.5	0.66	(0.29, 1.48)	120	111	92.5	33	26	78.8	0.30	(0.10, 0.88)
	5	133	111	83.5	52	41	78.9	0.74	(0.33, 1.66)	127	110	86.6	28	25	89.3	1.29	(0.35, 4.73)
	7	122	94	77.1	53	44	83.0	1.46	(0.63, 3.35)	99	89	89.9	32	27	84.4	0.61	(0.19, 1.93)
	9									86	69	80.2	29	23	79.3	0.94	(0.33, 2.68)
Grammar & punctuation[e]	3									120	106	88.3	33	27	81.8	0.59	(0.21, 1.69)

5	127	111	87.4	28	24	85.7	0.87	(0.27, 2.82)
7	99	84	84.9	32	22	68.8	0.39	(0.16, 0.99)
9	86	69	80.2	29	25	86.2	1.54	(0.47, 5.02)

[a] Reference group is children with isolated OFC. Children with an intellectual disability, children classified as exempt, and children who died before the year level test were excluded

[b, c, d] WALNA writing rubric changed in 2005. Results presented separately for periods 2000-04[b], 2001-04[c] and 2005-07[d]

[e] Spelling not tested at WALNA Year 9 level; Grammar & punctuation not tested in WALNA

CI, confidence interval; N, number participating; n, number reaching minimum standard; NAPLAN, National Assessment Program Literacy and Numeracy; OFC, orofacial cleft; OR, odds ratio; WALNA, Western Australian Literacy and Numeracy Program

Children with results over multiple school year levels in each domain showed high consistency in continuing to meet or not meet the national minimum standard (around 80-89% for WALNA, and 91-94% for NAPLAN assessments). The mixed logistic regression models for each program in each domain combined the records from around 3000-4800 tests over the four school year levels, for approximately 2100 children (Table 4). After adjusting for covariates, compared to children without OFC, children born with CLO were just as likely to meet the national minimum standard in all domains in each program (OR were less than 1, but 95%CI included unity), as were children born with CL+P (except for the NAPLAN spelling assessment). However, children born with CPO were less likely to meet the minimum standard in the NAPLAN reading and grammar and punctuation assessments, both numeracy assessments and the WALNA writing tests. In adjusted models, variables from the base model (child's sex, school year level, Indigenous background and socioeconomic quintile), were all significant.

Table 4. Odds ratios for meeting national minimum standards by test program and cleft type[a]

	WALNA				NAPLAN			
	Unadjusted model		Final model		Unadjusted model		Final model	
	OR	(95% CI)	OR	(95% CI)	OR	(95% CI)	OR	(95% CI)
READING								
CLO	0.89	(0.50, 1.59)	1.13	(0.63, 2.04)	0.91	(0.47, 1.77)	1.04	(0.51, 2.11)
CL+P	1.07	(0.61, 1.88)	1.29	(0.72, 2.32)	0.74	(0.39, 1.41)	1.04	(0.48, 2.26)
CPO	1.13	(0.72, 1.78)	0.88	(0.56, 1.39)	0.63	(0.40, 0.99)	0.57	(0.34, 0.96)
N children (N records)	*2147*	*(4397)*	*2120*	*(4337)*	*2108*	*(3188)*	*2094*	*(3171)*
FM adjusted for			*BM+child's relative age*				*BM+family order+child's relative age*	
NUMERACY								
CLO	0.75	(0.47, 1.20)	0.84	(0.51, 1.37)	0.74	(0.36, 1.52)	0.75	(0.36, 1.56)
CL+P	0.66	(0.43, 1.00)	0.71	(0.46, 1.11)	0.52	(0.28, 0.99)	0.73	(0.34, 1.57)
CPO	0.72	(0.49, 1.06)	0.64	(0.43, 0.95)	0.58	(0.35, 0.96)	0.47	(0.28, 0.82)
N children (N records)	*218*	*(4807)*	*2151*	*(4739)*	*2101*	*(3164)*	*2087*	*(3147)*
FM adjusted for			*BM*				*BM+school location*	
WRITING[b]								
CLO	0.70	(0.46, 1.08)	0.83	(0.53, 1.30)	0.66	(0.38, 1.15)	0.77	(0.39, 1.49)
CL+P	0.79	(0.52, 1.18)	0.88	(0.55, 1.39)	0.49	(0.29, 0.84)	0.77	(0.37, 1.59)
CPO	0.81	(0.58, 1.14)	0.66	(0.47, 0.92)	0.73	(0.48, 1.10)	0.62	(0.38, 1.00)
N children (N records)	*2163*	*(4742)*	*2134*	*(4674)*	*2108*	*(3193)*	*2094*	*(3177)*
FM adjusted for			*BM*				*BM+family order+birthweight*	
SPELLING								
CLO	0.78	(0.47, 1.30)	0.89	(0.50, 1.62)	0.78	(0.43, 1.43)	0.96	(0.49, 1.88)
CL+P	0.74	(0.46, 1.19)	0.88	(0.52, 1.48)	0.41	(0.25, 0.69)	0.52	(0.29, 0.94)
CPO	1.07	(0.70, 1.65)	0.94	(0.60, 1.46)	0.81	(0.53, 1.23)	0.68	(0.42, 1.10)
N children (N records)	*1903*	*(3346)*	*1879*	*(3300)*	*2112*	*(3203)*	*2098*	*(3186)*
FM adjusted for			*BM*				*BM+family order+school location+language background not English*	
GRAMMAR & PUNCTUATION								
CLO					0.78	(0.42, 1.44)	0.85	(0.43, 1.66)
CL+P					0.61	(0.36, 1.04)	0.92	(0.48, 1.79)
CPO					0.59	(0.39, 0.88)	0.49	(0.32, 0.76)

N children (N records)	*2112 (3203)*	*2098 (3186)*
FM adjusted for		*BM +family order+school location+birthweight*

[a] Reference group = children without OFC. Children with an intellectual disability, children classified as exempt, and children who died before the year level test were excluded

[b] WALNA writing - also adjusted for test period: ≤ 2004, 2005-07

BM, base model (variables included: OFC type, school year level, sex, Indigenous background yes/no, socioeconomic quintile), CI, confidence interval; CLO, cleft lip only; CL+P cleft lip and palate; CPO, cleft palate only; FM, final model; NAPLAN, National Assessment Program Literacy and Numeracy;OR, odds ratio; WALNA, Western Australian Literacy and Numeracy Program

School location: metropolitan, provincial, remote; family order: oldest or younger child in family; child's age relative to class: oldest, middle or youngest third; birthweight: <2500g or ≥2500g

Replacing the birth quintile of socioeconomic level with categories of parental education and occupation levels in NAPLAN data altered the estimate only for children with CPO in the reading assessment (adjusted OR 0.60 [95%CI 0.35, 1.05]); no other estimates changed. However, these variables included a category of unknown occupation and education level for 28% and 31% of children respectively. Time spent in hospital did not significantly alter the estimate for reaching the minimum standard for any domain.

DISCUSSION

Most children, born with OFC or not, met the national minimum standards. For most assessments, children with OFC had OR slightly but not significantly lower than children without OFC. However, estimates were significantly lower for children born with CPO in reading, numeracy, writing and grammar and punctuation assessments. Children born with CL+P had significantly lower odds for reaching the minimum standard in the NAPLAN spelling test, while children born with CLO had similar odds for reaching all minimum standards as children without OFC. This is the first population-based study of school test results for children with OFC that accounts for children with additional anomalies and suggests that children with OFC and additional major anomalies perform as well at school as children with isolated OFC.

Our results add support to the few population-based studies of school test results or intellectual capacity, where individuals with CPO consistently performed less well than individuals without OFC. Two Scandinavian studies of men at the time of compulsory military draft (~18 years), found that men born with CPO (but not men with CLO or CL+P) were more likely to record lower scores in intelligence testing than men without congenital

anomalies.[13 14] In these studies men with intellectual disability or other disability were excluded. Two studies have used linked school assessment data. In Iowa, tests completed by children with isolated CPO had lower mean percentile rankings in state-wide assessments over grades 2-11 in all subject areas. Tests completed by children with CLO ranked lower only for language, and tests completed by children with CL+P ranked lower for language and maths.[5] In Sweden, at the end of compulsory secondary school education, students born with CL+P or CPO were less likely to receive their Leaving Certificate and students with CPO were more likely to be in the lowest ranking levels for English and maths, but not students with CL+P or CLO for any subject.[6] Students with OFC attending public schools in Atlanta were 1.7-3 times more likely to be enrolled in special education services than children without congenital anomalies.[15] In contrast to our study, these school-based studies included children with isolated OFC, and inclusion of children with intellectual disabilities was not specified.

We could not adjust for some potential confounding factors. Maternal prenatal exposure to alcohol, tobacco and other drugs has been associated with poorer learning outcomes or school test results.[16-18] Psychosocial factors and mental and behavioral conditions may also adversely affect test scores,[19 20] but the relationship between these factors and OFC is unclear.[21] Breastfeeding has been associated with better overall achievement[22 23] and is known to be lower for children with OFC.[24-26] Neither could we account for the effects of home support or professional tutoring external to school. Members of CleftPALS WA report they make a concerted effort to ensure that their children are not disadvantaged by their OFC or by school absence related to OFC care. This effort may reduce the potential effects of OFC on school performance. In addition we could not adjust for within-school correlation, as individual schools could not be identified in our data.

School absence can affect school assessment scores[20 27] and effects may not be uniform across the curriculum, with writing affected more than reading and numeracy in NAPLAN tests[27] and maths being more vulnerable to absence than reading in standardised tests in Ireland.[20] Our use of hospital stay may be a poor proxy for school absence. Children with OFC, especially OFC involving the palate have outpatient care requirements (for speech therapy, dental and orthodontic appointments) that may be more likely to disrupt their school attendance than hospital admissions. However, absences for legitimate reasons (such as health care appointments or illness) have less influence on NAPLAN test results than unexplained absences or absences for reasons not acceptable to the school principal.[27]

Other factors associated with OFC, such as the high prevalence from a very young age of hearing difficulties secondary to otitis media, have been suggested as potential reasons for poorer school outcomes. While disparities in language development emerge early and can influence later educational outcomes,[28] the Longitudinal Study of Australian Children found that ear infections had minimal effect on vocabulary development from four up to eight years.[29] However, hearing difficulties may influence progress in other curriculum areas.

Many factors potentially affecting school test results are common to both children with CL+P and children with CPO (such as hospitalisations, middle ear conditions, outpatient health care needs)[30] and do not explain the differences in our results between children with these cleft types. Although having an additional anomaly did not affect estimates, the types of anomalies that occur with the different types of OFC vary.[31] Syndrome-like diagnoses are more common among children with CPO[31] and it is possible that at least some of those conditions, some of which may be unmeasured in our data, may predispose to learning impairment. In a

Norwegian cohort of children with OFC assessed at 10 years, additional conditions (including syndromes, developmental difficulties, attention and hyperactivity disorders and dyslexia), were more common among children with CPO (37%) than children with CL±P (22%). Children without these additional conditions had similar mean scores in cognitive functioning to the general population, but children with additional conditions had lower mean scores.[32]

Our linked data from many population based administrative data sets including a population-based congenital anomaly register with active surveillance enabled assessment of many factors potentially related to school test results, and ensured missing data were minimal. Completeness of registrations of individuals with OFC is estimated to be 100%[33] and with diagnoses up to six years of age, children with all cleft types were included. Linked data from the WA Data Linkage System have been validated and used extensively for health research and linkage proportions are >99%.[7]

While reaching the national minimum standards in these tests is not the only, nor necessarily the best measure of school achievement or ability, educational attainment is a major determinant of later success. Children not meeting these minimum standards are at risk of falling further behind, as without targeted intervention, they may not develop the required skills for progression through school. Children with OFC, particularly children with CPO, should be monitored to identify learning difficulties early, to enable intervention to maximise school attainment and longer term outcomes.

What was known

Some studies suggest that individuals with isolated orofacial clefts, especially those with cleft lip and palate (CL+P) or cleft palate only (CPO) have poorer school and cognitive outcomes. However there was no information about school performance for children born with orofacial clefts and additional anomalies, nor for Australian children born with orofacial clefts.

What this study adds

Most children met the national minimum standard in all subjects, whether they had an orofacial cleft or not. The proportion meeting the minimum standard was similar between children born with orofacial clefts with or without an additional major anomaly. Compared to children without orofacial clefts, children born with cleft lip only (CLO) were slightly less likely to meet the minimum standard in nearly all subjects (but this was not statistically significant). Children born with CL+P had the same results as children with CLO, except for one spelling assessment, where they were significantly less likely to meet the minimum standard. Children born with CPO were less likely to meet the minimum standard in several tests (NAPLAN reading, and grammar and punctuation, WALNA and NAPLAN numeracy, and WALNA writing).

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Contributors

JB, CR-G, CB and NN designed the study. JB, with support from RT analysed the data with methodological support from CR-G, CB, AD and NN. JB drafted the first version of the manuscript. All authors provided important intellectual content and gave their final approval of the version submitted for publication.

Competing Interests

None declared

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Ethics approval

The study protocol was approved by the WA Department of Health Human Research Ethics Committee and the WA Aboriginal Health Ethics Committee and recognised by the University of Sydney's Human Research Ethics Committee.

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