The Intellectual Function of Cleft Palate Children Compared on the Basis of Cleft Type And Sex

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Several studies of the intellectual function of children with different cleft types have been reported in the literature (*3, 5, 11, 12*). Studies employing the Stanford-Binet Intelligence Scale (SBIS) (*11, 12*) found no significant difference in measured intelligence between their cleft lip and palate (CLP) and their cleft palate only groups (CPO). Lewis (*11*) reported, however, that cleft lip and/or palate children with other anomalies had significantly lower intelligence quotients (IQ) than cleft lip and/or palate children without other anomalies.

In contrast to these findings, Goodstein (*5*) reported a significant difference in intellectual function between his CLP and CPO groups with the CPO group being significantly lower in all three Wechsler Intelligence Scale (WISC) IQs. Estes and Morris (*3*), in a similar study employing the WISC, reported a trend toward lowered scores for their CPO group in comparison with their CLP group. In addition, they also found no relationship between present hearing status, speech proficiency and IQ for their group. Thus, they concluded that the verbal IQ deficits found in cleft palate children must result from some factor or combination of factors other than poor hearing or poor speech skills. Lamb, Wilson and Leeper (*10*) came to the same conclusion regarding the effect of present hearing status on WISC verbal IQ after comparing normal hearing cleft lip and/or palate children with poor hearing cleft lip and/or palate children.

Cleft lip with or without cleft palate is considered to be genetically distinct from cleft palate only (*4, 21*). None of the studies (*3, 5, 11, 12*) which compared the intellectual function of the genetically distinct cleft types controlled their groups for age or sex composition. It would seem, in light of recent findings (*4, 13, 20, 21*), that differences in age and/or sex composition of the groups compared might influence results.

Nation (*13*), using the Peabody Picture Vocabulary Test (PPVT),

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found that normal hearing pre-school cleft palate children developed both comprehension and usage vocabulary at a slower rate than did their non-cleft siblings. If this finding can be generalized to older children, it would seem important to control for age in studies involving language abilities of cleft palate children.

Further, recent genetic studies (*20, 21*) have reported a possible interaction between sex and cleft type in which the sex having the lower population incidence of CLP or CPO would possess more of the polygenes necessary to produce the genetically distinct cleft types. This would mean that the female cleft lip and palate (FCLP) and the male cleft palate only (MCPO) would possess more of their respective cleft-producing polygenes than would their male cleft lip and palate (MCLP) and female cleft palate only (FCPO) counterparts since the FCLP and MCPO have the lower population incidence (*21*). According to Fraser (*4*), females with cleft lip and palate and males with cleft palate only may also have a greater number of other major malformations. Several authors (*4, 20, 21*) state that it is likely that a majority of both types of clefts have a polygenic mode of inheritance although they caution that, at present, there is much more evidence to support this hyopthesis in cleft lip and palate than there is in cleft palate only.

While Woolf (*20*) and Woolf, Woolf and Broadbent (*21*) state that the polygenically determined cases are those without other anomalies, Fraser (*4*) theorizes that both types of clefts could be part of a generalized developmental instability of a familial nature. This instability could also account for the increase in major malformations of other organs so often found in association with clefting. If Fraser (*4*) is correct, then the FCLP and MCPO groups might be expected to be more severely affected in other ways, perhaps in lowered intellectual function. Differences in the sex composition of compared groups of cleft children from study to study would then be a contributing factor in the failure to replicate results.

An additional contributing factor to the differences in results found in previous studies may have been the use of different test instruments. The two studies (*11, 12*) which employed the SBIS reported results in agreement with each other. Studies (*3, 5*) employing the WISC reported results similar to each other but in disagreement with the studies (*11, 12*) using the SBIS.

While previous studies of the intellectual function of cleft palate children (*3, 5, 11, 12*) did not consistently find significant differences in the intellectual function of the two cleft types, they did all conclude that cleft lip and/or palate children appeared to be lower in intellectual function than compared groups of non-cleft palate children. Further, the deficit was consistently found in the area of verbal intelligence.

The present study:

1. Examined a cleft lip and/or palate sample for differences in intellec-

tual function between two cleft types: Cleft lip with or without cleft palate (CLP) and cleft palate only (CPO).
2. Examined the hypothesis that there are sex and cleft type differences in intellectual function in which the FCLP and MCPO groups are significantly impaired intellectually in comparison to the MCLP and FCPO groups.
3. Examined the frequency of other anomalies in relation to sex and cleft type.
4. Examined the frequency of deviant hearing in relation to sex and cleft type.

Procedure

The sample was composed of 73 consecutive cleft lip and/or palate children between five and 16 years of age referred to the Cleft Palate Team of The Jewish Hospital of St. Louis.

Social classification of the group was determined through the use of Hollingshead's Two Factor Index of Social Position (*8*). None of the subjects fell into Class I, 8% were Class II, 14% were Class III, 38% fell into Class IV and 40% into Class V. On this index Class I is high, Class V is low. Most of the subjects were referred by Missouri Crippled Children's Service which handles families at the lower end of the socio-economic scale.

Subjects were divided into two cleft type groups using the classification system devised by Kernahan and Stark (*9*). The distinction between the two cleft types was made on the basis of presence or absence of a cleft of the lip and/or primary palate. The CLP group had either a cleft of the lip and/or primary palate with or without the presence of a cleft of the secondary palate. The CPO group had a cleft of the secondary palate only and no involvement of lip or primary palate. For a portion of the analysis, the two cleft type groups were further subdivided into male and female subjects within each cleft type.

The CLP group contained 44 subjects, 29 males and 15 females. There were three lip only males; two unilateral lip and primary palate, one male and one female; 11 complete bilateral lip and palate, eight males and three females; 28 complete unilaterals, 17 males and 11 females.

The CPO group contained 29 subjects, 16 males and 13 females. Of these, 21 subjects, 11 males and 10 females, had complete clefts of the secondary palate, while eight, five males and three females, had clefts of the soft palate only.

Three males and one female in the CLP group were black while one female CPO was black. One male CLP had lip pits, two male CPO children were Pierre Robin and one male CPO was Treacher-Collins. Information regarding the presence or absence of other anomalies was found in the medical records of the subjects. Hearing information reported was the

average decibel (dB) loss through the speech frequencies (250, 500, 1000 and 2000 Hz) re: ISO 1964 Standards). A pure tone air conduction hearing test was used. An average loss of less than 20 dB in the better ear was considered normal hearing while a loss of 20 dB or greater in the better ear was considered poor hearing. All 73 of the subjects were evaluated psychologically by the same examiner. Tests employed were the PPVT (*1*) and the WISC (*19*).

Seven comparisons were made in all. The first comparison was between the CLP and CPO groups without regard to sex. The subjects were then separated into four groups on the basis of sex and cleft type for the remaining six comparisons. The four resulting groups were male cleft lip and palate (MCLP), female cleft lip and palate (FCLP), male cleft palate only (MCPO) and female cleft palate only (FCPO).

Tests for unrelated samples (t tests) were used to compare the groups on the WISC and PPVT. The level of significance was set at .05 employing a one tailed test (*2*).

The data regarding hearing status and other anomalies were analyzed using 2 × 2 chi square designs for unrelated samples (*18*). The MCLP and FCPO groups were combined into one category while the FCLP and MCPO were grouped together to form a second category to obtain the required number of expected frequencies in each cell (*18*).

Results

A. Intellectual Function. Results of the comparison of the CLP and CPO groups without regard to sex are represented in Table 1. Data suggest no significant difference between groups on any variable except age. The CPO group was significantly older with a mean age of 11 years, while the CLP group had a mean age of 10 years.

Table 2 presents results of the comparisons of the four groups divided on the basis of cleft type and sex. There was no significant difference in

TABLE 1. Results of the comparison of the 44 cleft lip and palate (CLP) children with the 29 cleft palate only (CPO) children with no regard for sexual composition of the groups.

variables	*CLP group*		*CPO group*		*t*
	mean	*S.D.*	*mean*	*S.D.*	
SES	52	15	56	13	1.13
Age in mos	120	38	138	33	2.11*
PPVT IQ	91	20	92	21	.06
WISC verbal IQ	93	17	92	16	.30
WISC performance IQ	100	17	98	17	.44
WISC full scale IQ	96	17	94	17	.55

* Sig. <.05 two tailed; $df = 71$.

TABLE 2. Means, standard deviations and results of the comparisons made on the basis of sex and cleft type. $N = 73$

variables	*MCLP* ($n = 29$)	*FCLP* ($n = 15$)	*MCPO* ($n = 16$)	*FCPO* ($n = 13$)	*results*
SES $\bar{X}$	51	55	58	54	no significant differences between groups.
SD	17	11	11	15	
Age (mos) $\bar{X}$	115	129	146	128	difference between MCLP & MCPO: $t = 2.97^{***}$ $df = 43$
SD	35	44	31	34	no other significant difference
PPVT $\bar{X}$	95	85	88	96	groups compared / t / df
SD	19	21	18	24	MCLP-FCLP 1.59* 42
					MCLP-MCPO 1.08 43
					MCLP-FCPO .12 40
					FCLP-MCPO .53 29
					FCLP-FCPO 1.29 26
					MCPO-FCPO .93 27
WISC V $\bar{X}$	96	86	88	97	
SD	14	20	17	15	MCLP-FCLP 1.85** 42
					MCLP-MCPO 1.81** 43
					MCLP-FCPO .11 40
					FCLP-MCPO .17 29
					FCLP-FCPO 1.51* 26
					MCPO-FCPO 1.52* 27
WISC P $\bar{X}$	100	99	96	100	no significant differences between groups.
SD	18	17	18	17	
WISC FS $\bar{X}$	98	94	91	98	difference between MCLP and MCPO: $t = 1.32^{*}$
SD	16	18	17	16	$df = 43$

*** SIG. .01 (2 tailed)
** SIG. .05 (1 tailed)
* SIG. .10 (1 tailed)

socio-economic status (SES) between the four groups although the MCPO group was slightly higher in this regard than the other three groups.

The MCLP group, with a mean age of 10 years, was significantly younger than the MCPO group which had a mean age of 12 years. The FCLP and FCPO groups both had mean ages of 11 years. PPVT IQs were not significantly different for the four groups at the predicted level of significance but results were in the predicted direction. That is, the MCLP and FCPO groups earned higher scores than the FCLP and MCPO groups. The MCLP group earned a mean PPVT IQ of 95 while the FCLP

group earned a mean IQ of 85. On that same instrument, the FCPO group earned a mean IQ of 96 while the mean IQ of the MCPO group was 88. The difference between the MCLP and FCLP groups approached significance at the .05 level (1 tailed).

The WISC verbal IQ scores also followed the predicted direction with the differences favoring the MCLP and the FCPO groups. The mean WISC verbal IQ of 96 earned by the MCLP group was significantly higher (.05 level, 1 tailed) that the WISC verbal IQ of 86 earned by the FCLP group. The mean WISC verbal IQ of 97 earned by the FCPO group was not significantly higher than the mean WISC verbal IQ of 88 earned by the MCPO group at the predicted .05 level of significance but was significant at the .10 level (1 tailed). This was also true for the comparison of the FCLP and FCPO groups and the difference favored the FCPO group. The MCLP and FCPO groups appeared to be similar to one another on both PPVT and WISC verbal measures while the FCLP and MCPO groups also appeared to be similar to one another.

The mean WISC performance IQs were striking in their similarity for the four groups with a mean WISC performance IQ of 100 for the MCLP group, 99 for the FCLP group, 100 for the FCPO group and 96 for the MCPO group.

None of the mean WISC full scale IQs was significantly different from any other at the predicted level of significance. The differences were again, however, in the predicted direction and the difference between the MCLP and the MCPO groups was significant at the .10 level (1 tailed). The MCLP group earned a mean WISC IQ score of 98 and the FCPO group a score of 98 while the FCLP and MCPO groups earned scores of 94 and 91 respectively.

B. Hearing Loss and Other Anomalies. Table 3 shows the number and percentage of children with hearing loss >20 dB in the better ear as well as the number and percentage of children with other anomalies in each of the four groups. From information presented in Table 3, it appears that the FCPO group was least affected of the groups in the percentage of children with poor hearing and in the number of children with other anomalies.

TABLE 3. Number and percentages within the sex by cleft type groups with >20 dB average hearing loss in the better ear and number and percentages in each group having other anomalies.

sex × cleft type	*n*	*Ss w/>20 dB loss*		*Ss w/other anomalies*	
		n	*percentage*	*n*	*percentage*
MCLP	29	7	24	7	24
FCLP	15	6	40	5	33
MCPO	16	5	31	9	56
FCPO	13	1	8	1	8

TABLE 4. Results of the χ^2 for the combined categories: MCLP-FCPO vs. FCLP-MCPO on the variables of hearing status and presence-absence of other anomalies.

categories	*hearing status*			*other anomalies*		
	<20 dB loss	*>20 dB loss*	χ^2	*absence*	*presence*	χ^2
MCLP-FCPO	34	8	1.72*	34	8	4.06**
FCLP-MCPO	20	11	*df* = 1	17	14	*df* = 1

** Sig. .05 (1 tailed).
* Sig. .10 (1 tailed).

Only 8% of the children had a hearing loss and 8% had other anomalies. The MCLP group was next with 24% having poor hearing and 24% having other anomalies. The FCLP group had 40% with a >20 dB average hearing loss in the better ear and 33% with other anomalies, while the MCPO group had 31% with a >20 dB average hearing loss in the better ear and 56% with other anomalies. Results again followed the predicted direction with the MCLP and FCPO groups being less severely affected on the variables of hearing loss and other anomalies and the FCLP and the MCPO groups being more severely affected.

Table 4 shows results of the comparisons between the combined MCLP-FCPO group relative to the combined FCLP-MCPO group on the variables of hearing loss and presence of other anomalies. On the variable of normal hearing-poor hearing, results were not significant at the predicted .05 level but the difference was significant at the .10 level (1 tailed) with the MCLP-FCPO group having a smaller number of poor hearing children than the FCLP-MCPO group. The difference in the presence of other anomalies was significant at the .05 level (1 tailed) and again, the MCLP-FCPO group had fewer other anomalies than the FCLP-MCPO group.

Discussion

Data from Table 1 did not support findings from previous studies (*3, 5*) with which they would be most comparable. No significant difference in intellectual function between the two cleft types was found.

If the findings of Nation (*13*) in regard to the slower rate of language development in the cleft palate child can be generalized to older children, it may be that our CPO group had an advantage because they were significantly older. This does not seem likely, however, since further observation of the data from Table 2 indicated that the MCPO group was the oldest of the four groups as well as slightly higher in SES, yet they were the most impaired. In addition, previous studies (*3, 5, 11, 12*) may have had quite different sex compositions.

Data from Tables 2, 3 and 4 supported the hypothesis that there are

sex and cleft type differences with the sex having the lower population incidence of CLP and CPO being the more severely affected in verbal intelligence, in the presence of other anomalies, as well as in the frequency of poor hearing.

The differences found in the present study did not all meet the predicted level of significance, but did all follow the predicted direction. While verbal IQs of the MCLP and FCPO groups were only slightly lower than their performance IQs, the verbal IQs of the FCLP and MCPO groups were dramatically depressed in relation to their performance IQs. Further, in addition to having a greater percentage of children with poor hearing, the FCLP-MCPO group had a significantly greater number of other anomalies.

Data may lend support to the hypothesis of Fraser (*4*) that CLP and CPO are only one manifestation of a whole range of possible malformations due to a genetic developmental instability, and that the sex with the lower population incidence of CLP or CPO is more prone to concomitant physical anomalies in addition to a cleft of the lip and/or palate. Results were more favorable to this interpretation for CLP than for CPO. Findings were, to some extent, supportive of the earlier work of Lewis (*11*) who reported lower IQs for cleft lip and/or palate children with other anomalies.

It is of interest to note that information from Table 1 suggested that both the CLP and CPO groups were depressed in verbal abilities relative to their performance abilities. However, observation of data presented in Table 2 would suggest that it may be the more severely impaired FCLP and MCPO subgroups which contribute most to lowered verbal IQ scores for cleft lip and/or palate groups when compared to normal control groups. It may be that it is the FCLP and MCPO groups which are the language deficient groups and that the MCLP and FCPO groups are relatively normal in language skills. If this is true, further studies of the language function of cleft lip and/or palate children should be controlled not only for age, as Nation (*13*) suggests, but for sex and cleft type.

The fluctuating conductive hearing loss so commonly found in cleft lip and/or palate children (*6, 7, 14*) has often been considered a contributing factor to the verbal deficits consistently reported in these children (*3, 5, 10, 17*). The extent of the contribution of the hearing loss to the language deficit in cleft palate children is still unclear. The FCLP and MCPO groups had lower verbal IQs on both the WISC and PPVT. They also had the greater percentage of subjects with poor hearing. At the same time, some authors (*3, 10*) presently feel that hearing loss is not the only factor contributing to these verbal deficits. Lamb, Wilson and Leeper (*10*) found no difference in verbal IQ when cleft lip and/or palate children were divided on the basis of present hearing status using the same hearing criteria employed in this study. It may be fruitful to explore some of the other factors which could be contributing to these verbal deficits. One ap-

proach would be to investigate the possibility of a developmental anomaly of a central neurologic nature which could result in a deficit in language function. If cleft lip and/or palate is often associated with the malformation of other organs, this concept is certainly worth consideration in view of the consistent reports of language deficits among cleft lip and/or palate children. A battery of tests such as the Reitan Neuropsychological Test Battery for Adults and Children (*15*) could be used for this purpose. The Reitan battery (*15*), in addition to tapping verbal ability, tests differences in function on both sides of the body such as finger agnosia, speed of finger tapping and strength of grip which do not depend on past or present hearing status as a possible influence on performance.

Reitan (*16*) has studied brain-behavior relationships in children using this test battery (*15*). The WISC is included as part of the battery and has been found to be a powerful tool in discriminating brain-lesioned from non-brain lesioned children (*16*). Further research using a battery of tests such as Reitan (*15*) describes may help clarify the nature of the language deficits consistently reported for cleft lip and/or palate groups. It may be that the language deficit is more reflective of a developmental anomaly of a central neurological nature than it is of fluctuating conductive hearing loss.

The authors would be remiss if they did not point out that the auditory function of our subjects was tested at a stage well beyond the language acquisition age. To better understand hearing as a factor in depressed language function in cleft palate children, it would be essential to have a history of hearing status during the years when language is being acquired.

Summary

Seven comparisons of the intellectual function of cleft lip and palate (CLP) and cleft palate only (CPO) children were done employing the PPVT and the WISC. The first comparison was similar to previous studies (*3, 5*) in comparing a CLP with a CPO group without regard for the sex composition of the groups. The other six comparisons were made on the basis of sex and cleft type. In addition, hearing status and presence of other anomalies were reported for the four groups.

Results of the first portion of the study did not support results of previous studies (*3, 5*) which reported lower IQs for CPO groups when compared with CLP groups. Some of the possible reasons for differing results were discussed.

Information from the last six comparisons lent support to the idea of sex and cleft type differences. Data supported the hyopthesis of Fraser (*4*) who suggests that the more severe impairments may occur in the cleft palate person of the sex with the less frequent population incidence of cleft lip and/or palate, that is, the female cleft lip and palate (FCLP) and the male cleft palate only (MCPO) groups.

Results suggested that it may be the FCLP and MCPO groups who are

the language deficient subgroups of the cleft lip and/or palate groups and that language skills of the male cleft lip and palate (MCLP) and female cleft palate only (FCPO) groups are approximately normal.

A further exploration of sex and cleft type differences using an array of tests, such as the Reitan battery (*15*) was suggested in order to measure central nervous system function without involving past or present hearing status as a possible contributing factor to level of performance.

Finally, it was suggested that further studies of language function of cleft lip and/or palate groups be controlled for age, sex and cleft type.

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