Studies of Dysmorphogenesis in Children with Oral Clefts: II. Variant Palmar Patterning, Imperfect Ridge Formation and Increased Palmar Creasing as Indices of Dysmorphogenesis*

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Introduction

Previous studies have shown little or no association between oral clefting and variant dermatoglyphic patterns (1, 2). Such essentially negative findings are surprising in view of the frequently reported association of variant (though usually nonspecific) dermatoglyphic patterns with syndromes of multiple malformations (3). In the present investigation, we hypothesized that children with oral clefts, but without additional minor dysmorphogenetic features, would show relatively normal dermatoglyphic patterns, while those children with oral clefts plus additional minor features would also tend to show variant or unusual dermatoglyphic patterns. The reasoning behind this hypothesis has been presented in the first paper of this series (4).

Method

Complete palm-prints were available for study from 86 of the 101 schoolage children with oral clefts previously reported (4). "Variant" dermatoglyphic features were limited to those apparent on casual inspection. A total of nine such features were scored (see Table 1).

a. The axial triradius was classed as "distal" if located over 30% of the distance from wrist crease to base of 4th finger.

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	cleft palate			cleft lip-palate				combined				
	D_0	D_1	D_2	sum	D_0	D_1	D_2	sum	D_0	D_1	D_2	sum
number of individuals	4	7	21	32	18	16	20	54	22	23	41	86
1. missing digital triradius 2. missing or doubled	0	0	3	3	1	2	8	5	1	2	8	11
axial triradius	0	0	2	2	2	0	5	7	2	0	7	9
 distal axial tri- radius unusual palmar 	1	4	6	11	2	2	7	11	3	6	13	22
flexion crease	0	0	4	4	1	2	2	5	1	2	6	9
5. extra digital tri-				-				_				
radius.	0	1	0	1	2	0	3	5	2	1	3	6
6. thenar pattern 7. hypothenar pat-	0	2	5	7	4	1	4	9	4	3	9	16
tern	2	0	7	9	7	3	10	20	9	3	17	29
8. increased palmar creasing 9. variant ridge	0	0	3	3	1	0	3	4	1	0	6	7
formation	0	1	5	6	1	2	8	11	1	3	13	17
sum of variant find- ings	3	8	35	46	21	12	47	80	24	20	82	126

TABLE 1. Variant palmar patterning in relation to degree of dysmorphogenesis in children with oral clefts. D_0 , D_1 , and D_2 refer to zero, one and 2 or more dysmorphogenetic features respectively. Note that most variant dermatoglyphic features are more frequent in children with dysmorphogenesis (level D_2).

- b. The term "variant ridge formation" was used to refer to a variety of features:
 - (i) ridges interrupted by frequent gaps, producing a dashed-line or dotted-line effect.
 - (ii) deterioration of ridge pattern into a patternless array of dots.
 - (iii) presence of thin ridges lacking sweat pores.
 - (iv) multiple branching and island formation.

Examples of variant ridge formation are shown in Figure 1B.

Variant ridge formation was classed on a scale from zero to 3-plus. "Zero" and "trace" classifications were lumped as "absent"; 1-plus through 3-plus were lumped as "present".

c. Palmar creasing was graded on a scale of zero to 3-plus. A score of 2-plus or more was recorded as "increased palmar creasing".

Variant ridge formation and increased palmar creasing are obviously not variants of dermatoglyphic pattern per se, nor are variant palmar flexion creases. However, they are conveniently observed at the same time as the skin ridge patterns and are therefore conveniently included in studies of "palmar dermatoglyphics". The findings of the present study are not weakened by removal of these three factors from consideration.

Children had previously been classified by degree of dysmorphogenesis $(D_0, D_1, and D_2$ referring to zero, one and 2 or more minor dysmorphogenetic features respectively) and by school performance level (Level 1: A's, B's, and C's, Level 2: C's, and D's, one or more failures, held back a year; Level 3: in special education class or intellectually unable to attend school) For details of methodology, reference is made to the original report in this series (3).

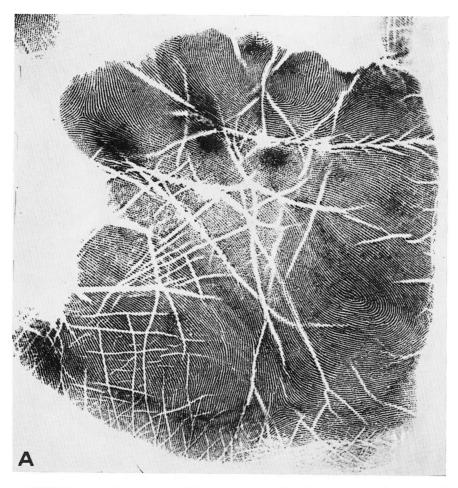


FIGURE 1. A. Palm print exhibiting pronounced palmar creasing but normal dermal ridge formation. B. Palm print exhibiting a high degree of imperfect ridge formation as well as moderate palmar creasing. Note particularly multiple branching and island formation in the hypothenar area, and (at the base of the 3rd digit and in the area of the axial triradius) multiple interruptions, producing dashed or dotted line effects.

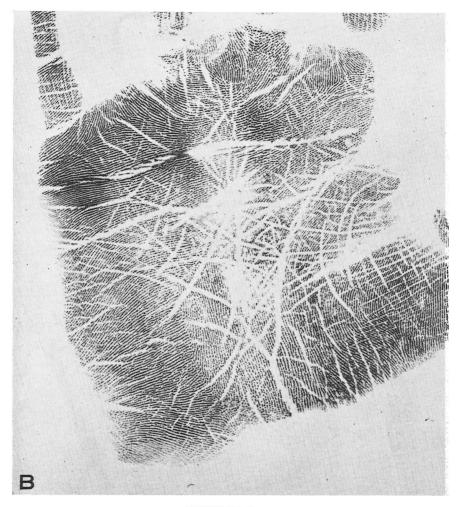


FIGURE 1 B.

Results

Table 1 demonstrates the generally higher frequency of individual variant dermatoglyphic traits in children with dysmorphogenesis (Level D₂). For example (bottom line of Table 1), a total of 126 variant features occurred in the 86 children tested, an average of 1.47 features per child or 0.74 features per hand. However, 82 of the 126 features occurred in the 41 children in level D₂, an average of 2.0 per child, as compared to 44 features in 45 children in levels D₀ & D₁, an average of about 1.0 per child. Thus, the overall frequency of variant features was twice as high in the dysmorphogenetic children. (χ^2 with 2 degrees of freedom for this comparison = 15.7, p \ll 0.005).

The effect appears to be stronger in children with CL(P), in whom the

	VP*	$\overrightarrow{VP}^{\dagger}$	sum	chi^2	p (two-tailed)
	<u> </u>		Cleft Palate	e	
D_{0-1}	8	3	11		
D_2	19	$\frac{2}{5}$	21		
sum	27	5	32	‡	p = 0.42 N.S.
		Cl	left Lip-Pal	ate	
D_{0-1}	23	11	34		
D_2	· 19	1	20		
sum	42	12	54	3.98	p < 0.05
		A	ll Oral Cle	fts	
D_{0-1}	31	14	45		
D_2	38	3	41		
sum	69	17	86	6.5	0.025

TABLE 2A. Association of variant palmar pattern and dysmorphogenesis in children with oral clefts. All variant features included. Note that children with dysmorphogenesis (level D_2) have higher prevalence of variant palmar dermatoglyphics.

* VP: Variant palmar pattern present.

 $\dagger \overline{\mathbf{VP}}$: Variant palmar pattern absent.

t p calculated by Fisher Exact Test.

frequency of variant features was 47/20 = 2.35 per person in level D₂, as compared to 35/21 = 1.7 for the corresponding group of children with cleft palate only. In both diagnostic categories, the frequency of variant features in level D₀₋₁ was about 1.0 per person. Probably because of small numbers, the trend does not reach statistical significance in the group with cleft palate alone.

Three of the nine variant features listed in Table 1 show no tendency to increased frequency in level D_2 . These are: extra digital triradius, thenar pattern, and hypothenar pattern. One is tempted to see in these three features the common trait of "added pattern" and to attribute to the remaining strictly dermatoglyphic features (1, 2, 3, possibly 9) the common trait of "reduced pattern". Removal of three "added pattern" features does not significantly alter the findings of Table 1 but greatly strengthens the association noted in Table 2.

In Table 2, the data are presented in terms of numbers of *individuals* with variant dermatoglyphic features, rather than in terms of numbers of variant *features*. The findings are similar to those of Table 1. Significantly larger numbers of children with variant features are found in level D_2 . In Table 2B the three variant features of "added pattern" have been omitted to produce a distribution based on "best" dermatoglyphic features.

The palms of several of the children exhibited what appeared to be increased wrinkling or crease formation. These are deep, sharply defined grooves in the palmar skin (distinct from the major flexion creases) which cut across the ridge patterns, interrupting them sharply like a knife cut.

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	VP	\overline{VP}	sum	chi^2	p (two-tailed)
			Cleft Palat	e	·
D_{0-1}	1	10	11		
D_2	13	8	21		
sum	14	18	32	6.2	0.01
		0	Cleft Lip-Pa	late	
D_{0-1}	10	24	34		
D_2	15	5	20		
sum	25	29	54	9.0	p < 0.005
			All Oral Cle	fts	
D_{0-1}	11	34	45		1
D_2	28	13	41		
sum	39	47	86	16.8	p < 0.001

TABLE 2B. Association of variant palmar pattern and dysmorphogenesis in children with oral clefts. Scoring based on "best" criteria (see text). Note improvement in significance levels (p-values) over those of Table 2A.

They tend to be straight or slightly curved, occasionally branching and when numerous, intersect and continue across each other at nearly right angles. They resemble the so-called "white lines" seen on fingerpads (5) and may possibly represent prenatal buckling or folding of the dermis due to 1) some difference from normal in the physical properties of connective tissue or 2) prenatal edema of the hands. Many, if not most, normal palms exhibit a few such creases. They are present in increased numbers in many but not all palms of children with Down's syndrome. Examples are given in Figure 1.

Of 7 children with increased palmar creasing, 6 exhibited 2 or more minor dysmorphogenetic features. In this series of 86 individuals, there was no clear tendency toward increase of palmar creasing with increasing age.

Discussion

Although previous reports have indicated little if any abnormality of the dermatoglyphics of children with oral clefts, the negative findings may have been due to failure to distinguish between children with and without additional minor dysmorphogenetic features. Such studies would ordinarily be "diluted" with nondysmorphogenetic children whose palm prints are apparently quite unremarkable.

The findings of the present study re-emphasize that variant dermatoglyphic patterns are apparently as a rule a function of generalized dysmorphogenetic processes rather than of specific congenital malformations (the exception being malformations of the hands themselves). In those cases in which an oral cleft is truly a localized phenomenon, there is no a *priori* reason to expect an unusual dermatoglyphic pattern. On the other hand, in those cases in which the oral cleft is only the salient manifestation of a generalized dysmorphogenetic process, one might expect that such a process could be reflected with some frequency in an unusual growth pattern of the hand and therefore, in an unusual dermatoglyphic pattern.

In the present series, the frequency of the "best" dermatoglyphic criteria was almost three times greater in children with two or more minor dysmorphogenetic features. Clinically detectable minor or major hand anomalies were present in about half of the children with variant palmar dermatoglyphics. We predict that quantitative hand pattern assessments ($\boldsymbol{6}$) will probably reveal subtle dysmorphogenetic changes in a high proportion of this sub-group of children with oral clefts, and that significant association between variant dermatoglyphic patterns of the "reduced pattern" variety and quantitative hand pattern changes will be found.

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