Numerical evaluation of facial pattern in children with isolated pulmonary stenosis

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SUMMARY A facial contouring technique, using light sectioning (Cobb, 1972), was modified (Ainsworth and Joseph, 1977) and used in a numerical study of children with isolated pulmonary stenosis (PS) to test the hypothesis that the facial pattern in this condition differs from the normal. Measurements were compared between a group of 20 normal children and a group of 20 children with PS between ages 6 and 10½ years. A distinctive facial pattern emerged. Many anteroposterior measurements were much greater in the PS group, indicating that the tissues were more prominent in the maxillary region. Altogether, 29 measurements showed significant differences ($P < 0.05$) between the two groups. Discriminant analyses were carried out to discover which, if any, might be used to predict the group to which an individual should belong. Depending on the variables chosen, between 34 and 37 individuals from the total of 40 were assigned to the correct group—PS or control.

Detailed measurements of facial abnormalities in the child have not been made. A general subjective approach has usually been used which relies on the experience of the observer. The lack of suitable measuring techniques has been responsible for this approach.

Facial patterns are common in some congenital heart diseases, notably supra-valvar aortic stenosis (Joseph *et al.*, 1966) and the cardiofacial syndrome (Cayler, 1969; Chantler and McEnery, 1971). It has been suggested that children with isolated pulmonary stenosis (PS) have moon faces (Wood, 1950), and Nadas (1963) referred to their round faces and broad cheeks. We too had a strong impression that patients with PS had a recognisable pattern and we wished to test this hypothesis by carrying out a detailed numerical analysis. An example of the facies in a child with PS is shown in Fig. 1.

The methods of moiré fringe contouring (Takasaki, 1970) and stereophotogrammetry (Ainsworth and Joseph, 1976) were considered but finally a light-sectioning device (Cobb, 1972) which produced facial contour maps was chosen. It was used after modifications had been made.

Fig. 1 Facies in a child aged 8 years with isolated pulmonary stenosis.

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Subjects and methods

Contour maps were made of the faces of 20 children (boys and girls) with PS, confirmed by cardiac

catheterisation or surgery. The patients came from Guy's and Brompton Hospitals. A control group of 20 normal children was obtained from Salomon's Centre, Guy's. Both groups of children were between ages 6 and 10½ years. The mean age of the PS group was 8 years (SD 1.66) and the mean age of the control group was 7.8 years (SD 1.42).

The contouring technique used has been described (Ainsworth and Joseph, 1977). Parallel slits of light were projected on to the subject's face, forming contours. They were photographed from a position perpendicular to their planes of projection. Plano-convex lenses, interposed in each optical path, prevented the light slits from diverging from their source and eliminated perspective distortion in the photograph. Full contour coverage of the face was achieved by rotating the head and taking multiple exposures. The exposures were orientated to a datum plane by an optical sighting system and linked by caliper measurements from life.

A life size contour map was drawn from each picture, producing a geometrically interrelated set of maps for each child. The sets of contour maps were processed by computer and sections of the face were drawn at the four levels, shown in Fig. 2, which also shows the landmarks used. The sections were

Table 1 *Measurements (A) of facial length, breadth, and height used for selecting the denominator of the facial index*

Measurements	Fig.	No.	Control		Pulmonary stenosis	
			Mean	SD	Mean	SD
Length	3	2	86.67	5.16	88.21	3.61
		63	74.50	3.85	76.25	4.02
Breadth	3	1	129.63	6.43	129.10	6.06
		41	124.90	6.73	124.30	6.03
Height	6	80	101.90	8.10	102.50	6.66

confined to the maxillary region because we suspected that the most characteristic features of the pattern were in this area.

Facial proportion index. The variation caused by differences in facial size between individuals was eliminated by calculating ratios (R) rather than by giving actual measurements. By comparing real, or absolute measurements (A), a suitable dimension was selected, and in each face the remaining measurements were expressed as percentages of this dimension. This provided a proportion index which produced ratio (R) values and enabled a more precise comparison to be made.

Initial investigation was confined to measurements (A) of facial length, breadth, and height (Table 1). Length was expressed by two measurements at different heights in the midsagittal plane (2 and 63). Breadth was expressed by the intertragal dimension (1) and the bizygomatic diameter (41), and height by the distance between the nasion and gnathion (80). The most reliable measurement for use as the denominator of the facial proportion index was the intertragal dimension (1), since it was measured by calipers from life between distinct landmarks.

Thus the facial proportion index is:

$$\frac{\text{absolute measurements (A)} \times 100}{\text{intertragal measurement } t^1 - t^2} = \text{ratio value (R)}$$

Results

Fig. 3 shows measurements 1–63 from which the mean values (R) for each group were used to construct composite sections at the levels (a), (b), (c), and (d). Broken lines represent the PS group, and continuous lines the control group (Fig. 4).

Sections (a). More measurements contributed to this pair of sections than to the other three pairs. Once the positions of the landmarks are established they remain common to all sections. It can be seen in Fig. 4 that the broken line is more prominent than the continuous one. In addition, in the PS group, the positions of both pairs of lateral and medial canthi

Fig. 2 *Landmarks and parallel planes of sections. Section (a) is defined by t^1 , sn , and t^2 . Section (b) is midway between section (a) and a horizontal line intersecting the canthi. Section (c) bisects the distance between section (b) and the canthi horizontal. Section (d) intersects (n).*

Fig. 3 Measurements in sections (1-63). Continuous lines indicate the distances measured. Where a line is both continuous and broken, the continuous part represents the distance measured. Values for these measurements are shown in Tables 2, 3, 4, and 5.

and the subnasal landmark are also more prominent and consistently offset to the left. In Table 2 measurement 5 is significantly greater in the PS than in the control group ($P < 0.001$) and so are 3, 16, 18, and 20 ($P < 0.01$), as well as 7, 22, and 23 ($P < 0.05$). Measurement 17 is significantly less in the PS than in the control group ($P < 0.05$).

The offset positions of the landmarks affect diagonal measurements originating from the tragi. Measurements 3, 5, and 7 from t^1 are significantly different between the PS and control groups because the landmarks are offset in the direction of the measurements. The opposite measurements 4, 6, and 8 from t^2 are not significantly different because the landmarks are offset approximately at right-angles to the direction of the measurements. Consequently, in the remaining sections the measurements were chosen carefully to exploit fully the differences observed. Therefore, the majority of diagonal measurements originate from or are related to t^1 .

Sections (b). The PS section is more prominent. In Table 3 measurement 28 is significantly greater in the

PS than in the group ($P < 0.01$) as are measurements 27, 30, 32, 33, and 38 ($P < 0.05$).

Section (c). The PS section is more prominent. However, the actual shape of the section, irrespective of prominence, is distinctly different at the side of the nose. The curve is more gradual compared with the sharper and more angular shape of the control section. In Table 4 measurements 43 and 54 are significantly greater in the PS than in the control group ($P < 0.01$) and so are 42, 44, 45, 47, 48, and 53 ($P < 0.05$).

Sections (d). Five measurements are sufficient to identify the sections above the upper eyelids and through the nasion. A curve was drawn through the co-ordinates of each section. The PS section is more prominent. In Table 5 measurement 59 is significantly greater in the PS than in the control group ($P < 0.01$) and also 61 ($P < 0.05$).

Asymmetry. Of the 25 significant measurements between the PS and control groups in the sections,

Fig. 4 Composite sections, viewed from above, which were constructed from the mean (R) values in each group. Broken lines show the shape of the face in the PS group, and continuous lines show the shape of the face in the control group. In section (a), points representing the positions of the four canthi in each group have been joined. Broken lines indicate the PS group and continuous lines the control group.

Table 2 Ratio values of measurements in section (a)
Fig. 3

No.	Control		Pulmonary stenosis		P
	Mean	SD	Mean	SD	
1	100.00		100.00		
2	66.89	2.97	68.46	2.83	
3	82.98	2.13	85.35	3.01	0.007
4	83.61	2.39	84.99	3.07	
5	47.55	1.71	50.20	2.18	<0.001
6	48.66	2.40	49.22	2.01	
7	61.25	1.83	62.95	2.42	0.018
8	61.65	2.33	61.83	2.14	
9	79.86	3.23	80.90	2.63	
10	79.15	3.17	80.51	2.55	
11	92.49	2.60	92.82	2.82	
12	92.53	2.67	92.24	1.92	
13	17.39	1.66	18.28	1.85	
14	17.49	1.37	18.17	1.69	
15	10.71	1.48	11.38	2.03	
16	10.69	1.38	12.22	1.82	0.005
17	108.90	5.20	105.00	4.84	0.023
18	52.37	2.51	55.30	3.16	0.003
19	53.25	2.78	55.98	2.84	
20	56.83	2.36	59.23	2.87	0.007
21	57.20	2.84	58.89	3.63	
22	59.59	2.34	61.90	3.16	0.013
23	59.73	2.68	61.84	2.68	0.019
24	43.46	1.86	43.99	1.60	
25	43.34	1.50	43.45	1.41	
26	93.14	1.72	93.11	1.70	

Table 3 Ratio values of measurements in section (b)
Fig. 3

No.	Control		Pulmonary stenosis		P
	Mean	SD	Mean	SD	
27	72.55	2.44	74.79	3.31	0.020
28	48.57	1.61	51.43	3.46	0.004
29	49.23	2.61	51.05	3.10	
30	53.65	2.77	56.36	3.73	0.014
31	53.68	2.88	55.45	3.20	
32	84.48	1.93	86.53	2.78	0.011
33	86.89	2.23	89.05	3.23	0.020
34	96.29	2.53	98.15	3.28	
35	7.42	1.22	7.87	0.90	
36	15.63	1.37	16.54	1.80	
37	16.07	1.99	16.72	2.12	
38	23.10	2.59	25.34	2.67	0.014
39	43.28	1.69	43.92	1.85	
40	43.78	1.70	43.29	2.20	
41	96.36	2.68	96.31	1.96	

there are 17 which have opposite measurements (not necessarily correspondingly significant) in the same relative positions on the other side of the face. Four of these are on the left side of the face (16, 23, 44, and 48), and 13 are on the right (3, 5, 7, 18, 20, 22, 28, 30, 43, 45, 47, 59, and 61). There are more significant

Table 4 Ratio values of measurements in section (c)
Fig. 3

No.	Control		Pulmonary stenosis		P
	Mean	SD	Mean	SD	
42	66.68	2.82	66.88	2.69	0.017
43	45.38	2.48	48.53	3.06	0.001
44	46.11	2.63	47.99	2.84	0.039
45	48.71	4.03	51.65	2.81	0.012
46	50.06	2.68	51.11	2.78	
47	52.46	2.97	54.64	3.27	0.036
48	52.23	2.62	54.29	2.64	0.026
49	6.69	1.90	5.95	0.92	
50	8.87	1.36	9.09	0.91	
51	13.09	1.54	13.90	1.68	
52	13.01	2.56	13.60	1.92	
53	19.30	2.32	21.07	2.19	0.019
54	57.25	1.96	59.56	2.88	0.006
55	82.96	2.08	84.34	3.08	
56	43.11	1.13	43.79	2.66	
57	42.81	1.70	42.65	2.26	
58	96.54	2.96	96.39	2.34	

Table 5 Ratio values of measurements in section (d)
and other regions

Region	Fig. No.	Control		Pulmonary stenosis		P		
		Mean	SD	Mean	SD			
Section (d)	3	59	47.53	2.39	50.27	2.60	0.002	
		60	48.15	2.89	48.96	2.55		
		61	50.43	2.82	52.35	2.58		0.033
		62	51.74	3.42	52.77	3.25		
		63	57.55	3.07	59.13	3.38		
Eyes	5	64	18.93	1.00	18.61	1.06	0.018	
		65	18.76	1.10	18.73	1.08		
		66	25.29	1.88	24.91	1.74		
		67	61.71	2.86	61.33	3.06		
		68	6.08	1.24	7.06	1.27		
		69	6.23	1.25	6.79	1.28		
		70	2.82	0.91	2.60	0.76		
		71	2.91	1.00	2.83	0.68		
Nose	5	72	23.17	1.78	24.44	1.61	0.023	
		73	31.91	2.12	32.87	2.36		
		74	32.96	2.09	34.16	2.02		
		75	32.73	1.83	34.08	2.69		
Nose	6	76	14.99	2.20	16.02	2.16	0.017	
		77	13.74	2.08	13.05	3.26		
Lateral	6	78	194.90	5.01	195.50	3.70	<0.001	
		79	86.99	2.83	85.84	1.88		
		80	78.52	3.74	79.74	4.72		
		81	53.38	1.60	56.10	1.97		
		82	66.10	1.63	67.72	2.28		

measurements on the right; this suggests that an asymmetrical pattern is present.

In each group asymmetry was measured by the differences between corresponding measurements on opposite sides of the face. The two most interesting pairs are 5 and 6, and 59 and 60, which both show differences at the 1% level between the two groups.

The opposite asymmetrical tendencies of both groups explain why, when coupled with greater anteroposterior measurements in the PS group, the majority of differences in the sections are on the right.

Eyes, nose, and side of the face. The values for

Fig. 5 Measurements relating to the eyes and nose (64–75). Values for these measurements are shown in Table 5.

Fig. 6 Lateral measurements in the midsagittal plane (76–80) and in 3 dimensions (81 and 82). Values for these measurements are shown in Table 5.

measurements associated with these components are shown in Table 5. Fig. 5 identifies measurements 64–75, and Fig. 6 identifies measurements 76–82. The height of the right palpebral fissure (68) is greater in the PS group ($P < 0.05$).

The maximum width of the lower portion of the nose between the alae (72) is greater in the PS than in the control group ($P < 0.05$). There is little difference in the length of the nose (73), although in the PS group it is slightly longer as are the other measurements of nasal length (74 and 75). Prominence of the tip of the nose (76) is marginally greater in the PS

group. The bridge of the nose at the nasion (77) is similar in both groups.

The maxillary angle (78) is similar in both groups. The facial angle (79) is particularly so. There is no difference in the measurement of facial height (80). The 3-dimensional distance between t^1 and ex^1 (81) is significantly greater in the PS than in the control group ($P < 0.001$). The 3-dimensional distance between t^1 and en^1 (82) is also significantly greater in the PS than in the control group ($P < 0.05$).

Statistical analysis. Of the 83 variables, 29 showed significant differences at the 5% level. This suggests that, for a particular individual, it may be possible to predict to which group, control or PS, he is likely to belong. The theory of discriminant analysis (Fisher, 1938; Rao, 1952; Hills, 1966; Armitage, 1971) gives rules for constructing a function of a set of variables, such that the sign (positive or negative) of the value of the function predicts the group (control or PS).

The best set of two variables consisted of 5 and 16. Their values are represented by points on a scatter diagram (Fig. 7). Points above the line correspond to positive values of the discriminant function—that is, individuals classified into the PS group. Four children with PS were misclassified as belonging to the control group, and one child was misclassified as belonging to the PS group.

The classification can be improved by using more

Fig. 7 Discrimination from two measurements between the group of control children (X) and group with PS (●).

variables. In Fig. 8 histograms for each group are shown for six variables.

The error rate of a discriminant function—that is the proportion of individuals it will misclassify—may be estimated by applying it to both groups of children. This will underestimate the error rate to be expected when used on other individuals, as the discriminant function has been chosen as the best for the sample groups. The estimate of Lachenbruch (1965) avoids this bias and gives a more accurate estimation. This is shown in Table 6 for several sets of variables. The error rate for six variables is now estimated as 4, as opposed to 2 in Fig. 8, where Lachenbruch's estimate was not applied.

Fig. 8 Histogram of discriminant function of variables 5, 16, 33, 44, 47, and 59.

Table 6 Error rates of the discriminant function

No. of variables	Delta Δ	Lachenbruch's estimate		Variables
		Proportion	Expected number of mis-classifications	
2	1.91	0.125	5	5, 16
5	2.68	0.15	6	5, 16, 33, 44, 47
5	2.65	0.15	6	7, 16, 68, 81, 82
6	2.93	0.1	4	5, 16, 33, 44, 47, 59
8	3.20	0.075	3	7, 16, 17, 30, 45, 53, 81, 82
10	3.65	0.075	3	5, 16, 17, 23, 28, 30, 33, 44, 47, 59

Discussion

Statistical analysis proves that children with PS have a distinctive facial pattern. Measurements can discriminate between both sample groups, classifying between 34 and 37 of the total of 40 children as either PS or control, according to the number of variables used.

All anteroposterior measurements in the sections, whether significant or not, are greater in the PS than in the control group. The majority emerge from a common source, a vertical plane coincident with the intertragal dimension t^1-t^2 (intertragal base plane).

In the PS group the face is longer from the intertragal base plane. A notable feature is the prominence of the subnasal and canthi landmarks, with correspondingly more prominent sections. However, the relationship between the forward landmarks in both groups, and the maxillary region considered in isolation without reference to the intertragal base plane, is much more subtle. For example, the positions of the four canthi, relative to each other in each group, are almost identical. Consequently, it is appropriate to consider the differences in two ways. Firstly, to devote special attention to the maxillary region, ignoring its relationship with the intertragal base plane, and secondly, to consider overall differences relative to the intertragal base plane which are responsible for the increase in facial length of the PS group.

Maxillary region. Comparisons can be made using the composite sections as a visual guide, but superimposing the canthi. The sections (a) for each group are different in shape. The PS section shows that the cheeks are slightly wider and more rounded. Measurement 16, which is highly significant, supports this and shows that there is a more prominent distribution of soft tissue in the cheeks continuing round to the sides of the nose adjacent to the alae. The position of the PS subnasal landmark (sn) is more prominent, and the angle $ex^1 sn ex^2$ (17) is less in the PS than in the control group, indicating more tissue at this point in front of the maxilla. This general 'bulge' becomes less pronounced in the other sections, but nevertheless is supported by two significant measurements, 38 in section (b) and 53 in section (c). The tissue covering the zygomatic bone resumes the same form as the control group.

In the region of the nose, the curve of the PS section (c) is more gradual than the steeper and more angular curve of the control section. Measurement 53 supports this. The sides of the nose near the bridge seem to be filled with excess soft tissue. This trend continues down the sides of the nose becoming

less and finally ending in a significant increase in the width of the nose between the wings (72). There is probably more cartilage at the sides of the nose and the tissue of the alae is more developed laterally.

Although the palpebral fissures are the same length and distance apart in both groups, the height of the right palpebral fissure (68), being significantly greater in the PS than in the control group, gives the eye a more rounded appearance with a slight decrease in height of the right upper lid. It is possible that a similar impression is produced in the left eye, but the actual measurements are not very different.

It can thus be seen that the differences in the maxillary region indicate different distributions of tissue in the PS group.

Anteroposterior differences. The much greater anteroposterior dimensions in the PS group could be caused by the maxillary region advancing anteriorly as a result of (1) excess tissue in that region only, or (2) excess tissue in the maxillary region, combined with a longer skull. Alternatively, the tragi may be situated more posteriorly as a result of (3) excessive posterior growth of the tragi, or (4) the complete ear is set back by a longer skull displacing the external auditory meatus. There could be a combination of these factors. However, it is clear that the differences are due either to soft or hard tissue, or to a combination of both. A definite conclusion, even with a detailed analysis of the skull, would be difficult to reach because different combinations of abnormal components can produce the same effect. Nevertheless, several points are worth noting.

If (1) is wholly or largely responsible, and the eyeballs remained in the same relative position to the intertragal base plane in both groups, the length of the palpebral fissures in the PS group would be very much less. The measurements confirm they are not. Therefore the eyeballs would have to be more forward in the orbits to bring their perimeters out to the more prominent positions of the PS canthi, which would be displaced anteriorly from the lacrimal and zygomatic bones due to a substantial increase in tissue around the eyes. This would mean that the eyeballs are being forced forwards in their orbits.

If (2) is responsible, the eyeballs would remain in the same relative positions to their orbits, and the canthi would be much the same distance from the bone in both groups.

It seems unlikely that (3) is involved to a large extent, as the ear would assume a very odd appearance which we did not observe.

It has been shown that a more prominent distribution of tissue in the maxillary region accounts for a portion of the increase in facial length in the PS

group. We think it is probable that, quite apart from altering the appearance and shape of the face in this region, an excess of tissue distributed throughout this area is wholly responsible for the greater anteroposterior measurements in the PS group.

Relationship to other organs. Since the face is repeatedly being looked at, the observer becomes skilled in discriminating between differing facial patterns. It is the subtle blending, structural form, relative sizes and shapes of the parts of the face which are responsible for differing facial patterns. In diseases not apparently affecting the form of the face, knowledge of its morphology may still be relevant. Thus it is conceivable that more subtle variations in the anatomy of the face are present which can be identified only by precise measurements. The face could be affected by some of the disturbances which have occurred in other organs during development.

In the normal developing fetus we do not know whether the embryological parts progress independently in an unrelated order, or fit into a precise system in an accurately timed sequence. If our tissues and organs do depend on one another for their origin and form, then we must base our observations on analysis of the embryo as a whole rather than restriction to individual organs. Any marked departure from normality in one organ might therefore be accompanied by maldevelopment in many other organs. Thus the facial pattern in children with PS may be an example of this interdependence of the form of various organs.

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