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Ear Morphology in Cleft Lip and Palate Anomaly

L. G. Farkas and W. K. Lindsay

Division of Experimental Surgery, The Research Institute and
Division of Plastic Surgery, Department of Surgery,
The Hospital for Sick Children, Toronto, Ontario, Canada

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Summary. The hyposthesis that the cleft lip/palate anomaly and the anomalies of the ears might be different manifestations of the First Arch Syndrome was tested. Five measurements and four qualitative signs recorded in 145 cleft lip/palate adults operated on in childhood were compared with similar data of 100 healthy Canadian males and females. The ears in clefts were significantly narrower and lower set with greater inclination of the ears' longitudinal axis to the facial profile line. In the frequency of the ear shape deformities there was no significant difference between the cleft patients and controls. The different cleft lip/palate forms revealed similar ear size and ear shape, not influenced by the extent of the cleft. Our study validates the hypothesis that in the case of growth disturbances of the face, qualitatively similar damages may be present in the auricles causing defects reminiscent of the early foetal condition regarded as a sign of some inhibition of growth and development.

Zusammenfassung. Die Hypothese, daß Lippen-, Kiefer-, Gaumenspalten und Anomalien der Ohrmuschel verschiedene Manifestationen von Entwicklungsstörungen im Bereich des ersten Kiemenbogens sind, wurde überprüft. Dabei wurden das Ergebnis von 5 Messungen sowie 4 qualitative Zeichen bei 145 Erwachsenen, die in der Kindheit an Lippen-, Kiefer-, Gaumenpalten operiert waren, mit den Daten von 100 gesunden kanadischen Männern und Frauen verglichen. Die Ohrmuscheln der Spaltenpatienten saßen deutlich tiefer und näher am Kopf mit einer größeren Neigung der Ohrlängsachse zur Profillinie des Gesichts. Verformungen der Ohrmuschel wurden dagegen in beiden Kollektiven gleichermaßen gefunden. Die Veränderungen der Ohrmuschel waren unabhängig von der Größe der Spaltbildung. Die Untersuchungen unterstützen die Hypothese, daß bei Wachstumsstörungen des Gesichts bleibende Defekte an der Ohrmuschel vorhanden sein können als Zeichen einer Behinderung von Wachstum und Entwicklung.

An abnormally shaped or abnormally sized auricle is a true congenital malformation occurring in association with other malformations sometimes as a sign of a generically determined syndrome (Burian, 1954; McKenzie and Craig, 1955; Stiegler and Berry, 1958; Green, 1963; Kraus *et al.*, 1963; Gorlin and Pindborg, 1964; Hilson, 1966; McKenzie, 1966, 1968, 1971; Fara *et al.*, 1967; Longacre, 1968; Konigsmark, 1970; Leiber, 1972). Anomalies of the auricle frequently accompany anomalies of the genito-urinary tract (Potter, 1946; Hilson, 1957; Longenecker *et al.*, 1965; Taylor, 1965).

Anomalies of the auricles have been found in more than one-third of a series of one thousand cleft lip/palate patients (Farkas *et al.*, 1970). Hajnis and Farkas (1968) also found a significantly higher frequency of abnormally shaped and sized auricles in 110 young cleft lip/palate patients than in controls.

Since minor structural anomalies of the auricles might be helpful indicators of altered embryonic development (Smith and Bostian, 1964; Leiber, 1972), we studied the frequency of auricular deformities in adults born with different forms of cleft lip and palate anomaly in whom development of the auricle was now complete.

Material and Methods

Our study consisted of 145 young adults of both sexes between the ages of 16 and 20 years, who had undergone surgical repair of cleft lip/palate before two years of age at the Hospital for Sick Children, Toronto, and were available for re-examination. Twenty-nine had had bilateral cleft lip and palate, 74 unilateral cleft lip and palate and 42 isolated cleft palate, among them 56 incomplete and 89 complete clefts. All cleft lips had been repaired by LeMesurier's operation and the cleft palates by a modified Dorrance pushback operation. No surgery has been performed on the ears.

Both auricles were measured and various qualitative signs recorded. Values obtained were compared to those of a control group of like age made up of 50 Canadian males and 50 females, all healthy Caucasian volunteers. Measurements of subjects and controls were made by one of the authors (L. G. Farkas).

Five measurements of each auricle, some of them modified from those used in standard anthropometry (Martin and Saller, 1957; Hertzberg *et al.*, 1963; Hajnis and Farkas, 1969; Weiner and Lourie, 1969) were made directly on patients and four qualitative signs for each auricle were recorded.

Measurements

1. Auricle Length. The maximum length of the ear measured vertically from the lowest point of the lobe to the highest point of the helix (Fig. 1).

2. Auricle Breadth. The maximum width of the ear measured by sliding caliper: the width is the greatest distance between a line joining the superior and inferior attachements of the auricle and the posterior border of the helix (Fig. 1).

3. Auricular Protrusion. The angle between the dorsal surface of the upper helical margin of the auricle and the plane of the pars petrosa of the os temporale measured by transparent protractor and expressed in degrees. Holding the protractor horizontally with its zero mark above the upper insertion of the auricle (otobasion superius), the straight side of the protractor is pressed firmly to the head and the size of the angle noted. The auricle was classified as protruding when the angle was 40° or more (Fig. 2).

4. Auricular Inclinaton. The angle between the longitudinal medial axis of the auricle and a line through the external auditory meatus parallel to a line joining the supraorbital prominence and the menton. The angle is measured by transparent protractor (Fig. 3).

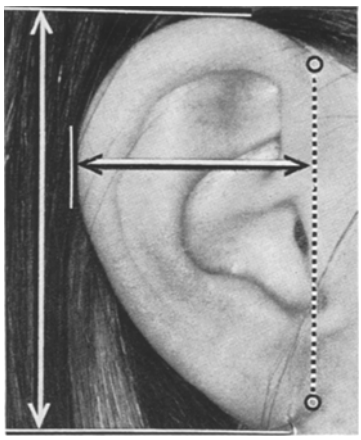

Fig. 1. Photograph showing measurement of the length and width of the auricle

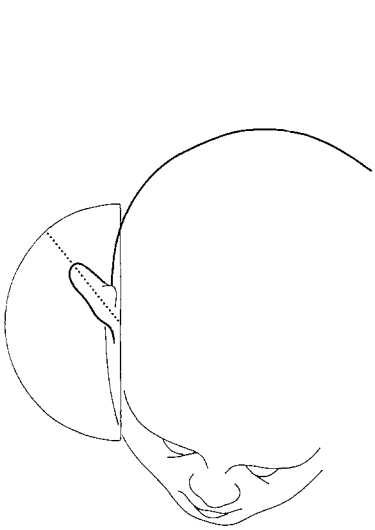

Fig. 2

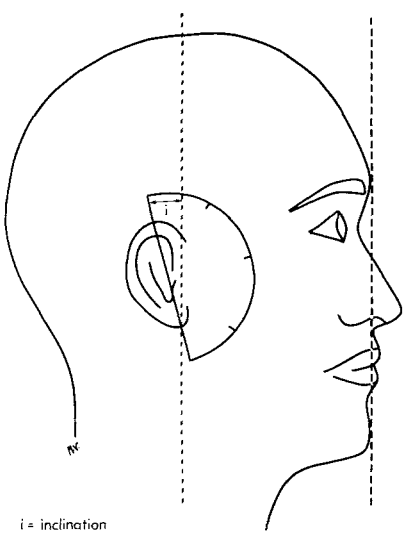

Fig. 3

Fig. 2. Diagram illustrating method of measuring auricular protrusion. A transparent protractor with its base pressed firmly against the side of the head is placed at the level of the tip of the auricle with the zero mark above the otobasion superius. The angulation of the auricle is read in degrees

Fig. 3. Diagram. The angle of inclination is assessed by placing the protractor with its base along the longitudinal axis of the auricle. The angle between the long axis of the auricle and a line parallel to a line joining the supraorbital prominence and the menton is read in degrees

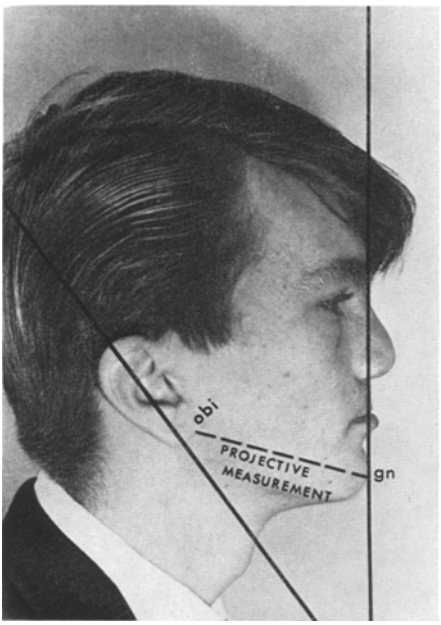

Fig. 4. Photograph showing measurement of auricular insertion. The direct distance between the otobasion inferius (*obi*) and the gnathion (*gn*) is measured by spreading caliper. The auricle pictured is low set. The tip of the auricle should be on a level with the eyebrow and inclined 24° C

5. *The position of the lower insertion of the auricle* in relation to the menton (Figalova and Farkas, 1968): The straight line distance between the inferior attachment of the auricle (*obi*, otobasion inferius) to the face and the gnathion landmark (*gn*) was measured by spreading caliper (Fig. 4).

Qualitative Signs

1. *General configuration of the auricles* was stated as normal or abnormal. The abormal auricle was Macacus (Fig. 5) or Cercopithecus-type, or having a concave abnormality of the upper lateral helical border.

2. *The helix* was described as flat unfolded helix (Fig. 6); slightly rolled narrow helix; moderately rolled normal helix; markedly rolled wider helix; and wide helix covering the scapha (Hajnis and Dobisikova, 1968; Weiner and Lourie, 1969) (Fig. 7).

3. *The lobe* was classified as attached if its lowest point was attached to the cheek (Fig. 7), or as free, regardless of the degree of freedom (Lai and Walsh, 1966; Hajnis and Dobisikova, 1968). Asymmetrical earlobe attachment was analyzed for this particular sign only.

4. *The lobe size* was classified as hypoplastic (Fig. 7) or well developed and as medium, large, or very large (Weiner and Lourie, 1969). For statistical evaluation of results chiquadrate Student's *t*-test and the statistical analysis of the significance of the observed differences (Hughes, 1969) were applied.

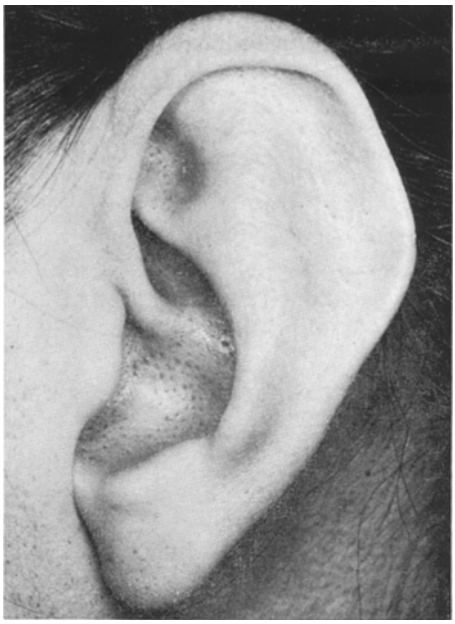

Fig. 5. Macacus-type auricle. The general shape of the posterior margin is sharply angulated

Fig. 6

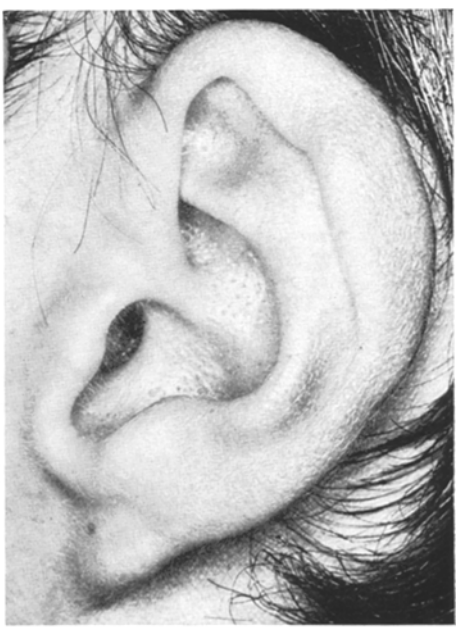

Fig. 7

Fig. 6. Flat helix
Fig. 7. Wide helix covering the scapha. Hypoplastic attached earlobe

Results

A. Measurements

1. The Whole Study Group

Both auricles of male patients and the left auricle of female patients were significantly *narrower* than those of controls (Table 1). The *length* of the auricles in patients was smaller than in controls but only in males was the difference significant. In all patients the auricles were significantly *lower set* than in normals with the exception of the right auricle in males where the difference was of borderline significance (Table 2).

Auricular protrusion in male patients ranged from 20 to 45 degrees, in female patients from 14 to 40 degrees. The incidence of protruded auricles, 40° or more, was similar in patients, 9 of 145 cases, and controls, 5 of 100 patients.

The *inclination* of the longitudinal axis of the auricles in patients ranged from 10 to 45 degrees (Fig. 4), in controls from 2 to 30 degrees. The inclination in those with unilateral cleft lip/palates, bilateral cleft lip/palates and isolated cleft palates was significantly greater than in controls ($P < 0.05 - P < 0.001$).

2. Unilateral Cleft Lip and Palate

The *width* and the *length* of the auricles in male patients (49) was significantly less than in normals, both on the cleft and non-cleft side ($P > 0.001$) while in female patients (24) the differences were not significant. There was no difference between the size of the auricle on the cleft and the non-cleft side in our study group.

Also the auricles of patients were lower set than in controls when assessed by the direct distance measurement of the lower insertion landmark from the gnathion but only in female patients was the difference significant ($P < 0.001$).

3. Bilateral Cleft Lip and Palate

The auricles of patients were significantly narrower ($P < 0.001$) but no shorter than those of controls. In the direct distance of the lower insertion points from the gnathion there was no statistical difference between the patients and the normals.

4. Isolated Cleft Palate

The auricles of patients with isolated cleft palate were significantly *narrower* ($P < 0.05 - P < 0.001$) than the ears of controls, and shorter but not to a significant degree.

The auricles of the patients were significantly *lower set* ($P < 0.001$) than those of controls.

Table 1. Measurements of the auricles in patients with cleft lip/palate and controls

Measurements	Sex	Cleft group ($N = 144$)			Controls ($N = 100$)			Differences (t-test)
		No.	Mean	SD	No.	Mean	SD	
Length of auricle	M	90	61.4	4.5	50	63.4	3.4	$T = 2.7386$
								$P < 0.01$
—Right (sa-sba)	F	54	57.7	2.8	50	58.4	3.3	not significant
Length of auricle	M	90	61.7	4.2	50	64.5	3.6	$T = 2.9733$
								$P < 0.01$
—Left (sa-sba)	F	54	58.2	2.13	50	59.2	3.2	not significant
Width of auricle	M	90	34.9	2.3	50	37.1	2.4	$T = 5.3423$
								$P < 0.001$
—Right (pra-pa)	F	54	33.1	2.5	50	33.4	1.9	not significant
Width of auricle	M	90	34.5	2.2	50	36.5	2.6	$T = 4.8285$
								$P < 0.001$
—Left (pra-pa)	F	54	32.1	2.1	50	33.2	2.1	$T = 2.6699$
								$P < 0.01$

Table 2. Measurements of the position of the auricles on the head in cleft lip/palate group in comparison with controls

Measurements	Sex	Study group ($N = 81$)			Controls ($N = 100$)			Differences (t-test)
		No.	Mean	SD	No.	Mean	SD	
Direct distance (gn-obi)	M	45	117.1	6.7	50	119.2	5.1	$T = 1.7296$
								$P > 0.05$ (not significant)
right	F	36	109.0	8.9	50	112.8	4.9	$T = 2.5377$
								$P < 0.02$ (significant)
Direct distance (gn-obi)	M	45	117.2	7.2	50	119.9	4.8	$T = 2.1709$
								$P < 0.05$ (significant)
left	F	36	107.2	8.4	50	113.6	5.2	$T = 4.3602$
								$P < 0.001$ (significant)

Table 3. The shape deformities in the study group in controls in comparison with the data from literature

Qualitative signs	Study group ($N = 145$)	Controls ($N = 100$)	Data from the literature (normals)
Normal ear form	82 (56.5%)	63	
Helix deformities (unilateral or bilateral)			
Unfolded (flat helix) (Fig. 6)	2 (1.3%)	2	From 0.47% to 17% (Geyer, 1928; Hajniš and Dobisikova, 1968)
Flat helix with total ear form anomaly (Cercopithecus and Macacus type) (Fig. 5)	1 (0.6%)	1	From 1% to 20.6% (Vali, 1893; Schwalbe, 1897; Hajniš and Dobisikova, 1968)
Narrow slightly rolled helix	3 (2%)	2	25.7% in males, 5.71% in females (Hajniš and Dobisikova, 1968)
Markedly rolled wide helix	16 (11%)	9	17.1% in males, 20% in females (Hajniš and Dobisikova, 1968)
Wide helix covering scapha (Fig. 7)	17 (11.7%)	9	From 0.7% to 28.5% (Schwalbe, 1897; Geyer, 1936; Rysanek, 1949, 1950; Lange, 1966; Hajniš and Dobisikova, 1968)
Concave abnormality of ear contour	7 (4.8%)	—	
Ear lobe deformities			
Unilateral attached ear lobe (Fig. 7)	4 (2.7%)	8	5% (Hajniš and Dobisikova, 1968)
Small (hypoplastic) ear lobe (Fig. 7)	13 (8.9%)	6	48.5% in males, 54.2% in females (Hajnis and Dobisikova, 1968)

B. Qualitative Signs

There was an insignificant difference in the total number of auricular shape deformities between the study group, 63 of 145 cases, and the controls, 37 of 100 cases (Table 3). The concave abnormality of the helical margin of the auricule was noted in 7 cleft lip/palate patients and in no controls. Auricular deformities in the study group were seen insignificantly more often bilaterally, 36 of 145 cases, than unilaterally, 27 of 145 cases. A unilaterally deformed auricle was recorded significantly more often on the cleft side, six cases, in unilateral cleft lip/palate forms than on the non-cleft side, two cases. The frequency of auricular deformities was independent of the type of cleft lip/palate deformity.

C. Relationship between the Extent of Cleft ing and Auricular Abnormalities

There was no statistically significant difference between auricular size and form in patients with incomplete ($N = 55$) and those with complete cleft lip/palate ($N = 89$).

Discussion

We used standard anthropometric measurements of the auricles in cleft lip/palate patients and added assessment of the position of the auricles relative to the head. We developed a relatively simple method of measuring the inclination of the auricles based on the so-called general profile line used by plastic surgeons to measure the protrusion of the nose (Baud, 1966; Núñez, 1970). In many instances malposition was the only auricular sign of congenital damage extending below the area of the middle face.

The high incidence of auricular deformities associated with cleft lip/palate has been noted by many authors (Burian, 1954; Stiegler and Berry, 1958; McKenzie, 1966; Bardach, 1967; Hajnis and Farkas, 1968; Farkas *et al.*, 1970), although what constitutes a deformity is difficult to define. Some may be abberations within the normal range (Mustardé, 1971). Comparisons are difficult because methods of measurement differ (Churchil and Truett, 1957; Smith, 1970; Davis, 1971; Kleinfeldt and Dahl, 1971; Leiber, 1972). Also the frequency of the subjective signs differ from population to population (Schaeffer, 1892, 1893; Pöch, 1926; Hildén, 1935; Vassal, 1954; Jürgens *et al.*, 1962; Dronamraju, 1966).

As did Hajnis and Farkas (1968) in a study of 110 young Czech cleft lip/palate patients, we found that the width of the auricle in cleft patients was less than that in controls.

It is beyond the scope of this paper to decide whether *all* deformities of the auricle found in cleft lip/palate patients are truely congenital anomalies. We regard the flat, unfolded helix as an anomaly. In our

patients it occurred singly or in combination with a change, either of the Macacus or Cercopithecus type, in the total ear. The enrolling process of the helix usually takes place between the fourth and eighth month of intrauterine life (Stanek, 1952).

We classified the concave abnormality of the auricular contour seen in seven cleft lip/palate patients as a congenital anomaly as did Hajnis and Farkas (1968). True auricular anomalies associated with cleft lip/palate may be explained by interactions between two organizer centers of the face as noted in Pfeifer's hypothesis (1967), or they may be regarded as different manifestations of the First Arch Syndrome according to McKenzie's hypothesis (1968).

The low placement of the auricles seen in our patients was also reported by Smith and Bostian (1964) in two cleft lip/palate patients with mental retardation. Malposition of the auricles may result from defective development of the middle and lower face (Johnston). The facial tissue deficiencies in front of the auricle, especially in the mandibular region, caused by primary failure of the cranial neural crest, may secondarily influence the position of the auricle. The auricles themselves, formed largely from mesoderm, can be relatively unaffected (Johnston). However, the extremely low set auricle with its more oblique longitudinal axis, reminiscent of the auricle in early foetal life (Caronni, 1971), may indicate some inhibition of growth and development (Rogers, 1968; McKenzie, 1971).

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L. G. Farkas, M.D., F.R.C.S.(C), D.Sc.
555 University Avenne
Toronto, Ontario
Canada