Anthropometry of the Face in Lateral Facial Dysplasia: the Unilateral Form

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The *skull, face* and *ears* of 46 patients with the *unilateral* form of *lateral facial dysplasia* were examined at the Hospital for Sick Children, Toronto, using *anthropometric methods* for the purpose of contributing to the morphology of this syndrome.

The classical picture of unilateral facial asymmetry with microtia was found in 13 cases (28.3%).

The study showed bilaterally defective mandibular arc measurements in 28 per cent of the cases. The facial damage was associated in 15 per cent with ears normal in size but defective in location, level, and shape.

The anthropometric examination revealed a great variety of defects on both sides of the face, often barely detectable by routine inspection. It was also found that great deficiencies in the surface measurements were accompanied by great bone deficiencies.

The value of measuring the soft tissues of the anomalous face by x-ray cephalometry is limited, and subjective visual evaluation is inaccurate. In morphological studies of the face, cephalometry applying surface measurements of the face and head yield helpful information (Farkas et al., 1966; Farkas and Lindsay, 1973b; Fraser and Pashayan, 1970; Hajniš and Figalová, 1973; Peyton and Ritchie, 1936; Williams, 1968).

Lateral facial dysplasia is synonymous with microsomia (Converse et al., 1973; Gorlin anf Pindborg, 1964), auriculo-branchiogenic dysplasia (Caronni, 1971) and otomandibular dysostosis (Francois, 1961), all of which are basically first and second branchial arch syndromes (Grabb, 1965). The term used by us (Ross, 1975b) indicates that the anatomical changes are not exclusively on one side of the face.

The purpose of this study was to establish the main characteristics of the skull, face, and ear in the unilateral form of lateral facial dysplasia (LFG) using anthropometry.

Material and methods

Forty-six white patients, 29 males and 17 females, between four and 16 years of age were examined at the Hospital for Sick Children, Toronto. All had unilateral LFD diagnosed by clinical and roentgenographic examination (Ross, 1975b). Surgical repair, three for macrostomias and 37 for microtias, had not substantially altered the original shape of the face and head.

Thirty-three surface measurements were made and 16 qualitative signs (Farkas and Lindsay, 1971; Farkas and Lindsay, 1972; Farkas and Lindsay, 1973a; Lindsay and Farkas, 1972; Martin and Saller, 1957; Weiner and Lourie, 1969) were noted for the face of each patient. The study was

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completed by roentgenological examination of the mandible (Ross, 1975a) and temporal bone (Harwood-Nash, 1974).

The surface measurements were compared to similar data from healthy West German boys and girls of corresponding age (Hajniš, 1974). Statistical analysis of the material was completed by using standard methods. Measurements differing by ±2 SD or more from the normal values were designated as "abnormally" short or long.

Results

Neurocranium

Abnormal measurements. Fourteen out of 46 patients had at least one abnormal measurement: a short or long skull base width (t-t), a short skull length (g-op), or a short skull width (eu-eu) (Figure 1).

Face

Abnormal vertical measurements. Height of the upper face (n-sto) was long in eight patients. Height of the lower third of the face (sto-gn) was short in five patients, and total facial height (n-gn) was long in four patients (Figure 1).

Abnormal horizontal measurements. Tragion-gnathion (surface) half-arc (t-gn) was short in 37 patients. Tragion-subnasale (surface) half-arc (t-sn) was short in 26 patients, and the bizygomatic diameter (zy-zy) was short in 11 patients (Figure 2).

The bitragion-subnasale arc generally covers the maxillary region, while the bitragion-gnathion arc corresponds to the mandibular region.

Figure 3 shows the various combinations of short and normal arc measurements in the middle and lower third of the face. Short maxillary and mandibular half-arcs on the same side of the face were noted in 18 patients. In five patients, both halves of the maxillary and mandibular arcs were short. Normal arc measurements were found in both maxillary and mandibular regions in 16 patients.

Asymmetry in the maxillary and mandibular arcs. The asymmetry found between normal half-arcs of the maxilla and mandible was of slight degree (2–4 mm), a routine finding in the normal population. The average asymmetry increased when both half-arcs were short and reached the maximum when only one half-arc of the maxilla or mandible was short in the maxilla, 15.3 mm and, in the mandible, 15.2 mm.

In almost two-thirds of the patients (11 out of 17) with at least one short mandibular half-arc and marked asymmetry of 10 mm or more between the mandibular half-arcs, roentgenograms revealed severe asymmetry in the mandibular length and deficiencies in the temporamandibular joint (Ross, 1975a). On the other hand, roentgenographic examination revealed a mild defect of the mandible in six out of 19

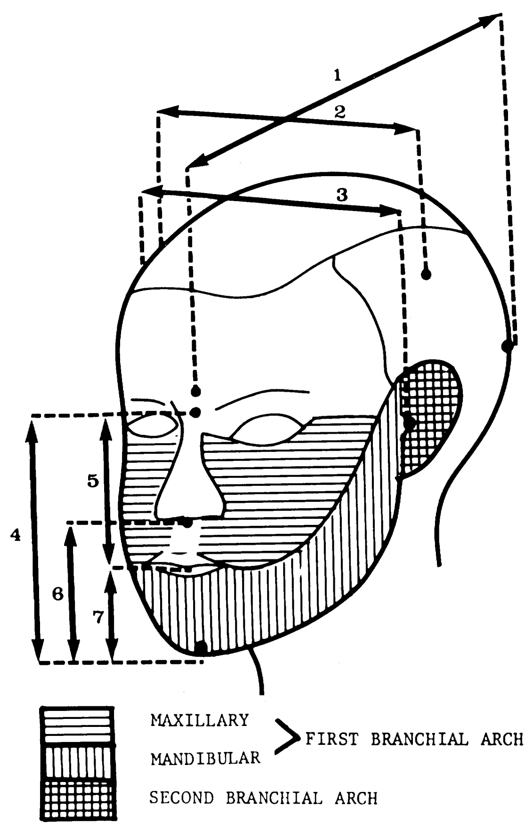

FIGURE 1. Measurements of the skull and face schematically. 1 = length of the skull (g-op = glabella-opisthion). 2 = width of the skull (eu-eu = euryon-euryon). 3 = width of the skull base (t-t = tragion-tragion). 4 = total facial height (n-gn = nasion-gnathion). 5 = upper face height (n-sto = nasion-stomion). 6 = lower face height (sn-gn = subnasale-gnathion). 7 = height of lower third of the face (sto-gn = stomion-gnathion).

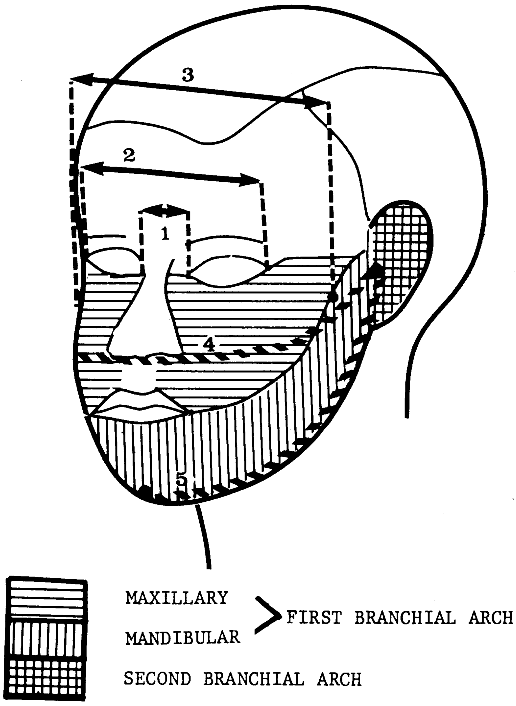

FIGURE 2. Measurements of the face schematically. 1 = interocular diameter (en-en = endocanthion-endocanthion). 2 = biocular diameter (ex-ex = exocanthion-exocanthion). 3 = bizygion diameter (zy-zy = zygion-zygion). 4 = maxillary arc (t-sn-t = tragion-subnasale-tragion). 5 = mandibular arc (t-gn-t = tragion-gnathion-tragion).

patients who had a slight difference between the normal half-arcs of the mandible.

In seven patients, the face was found to be symmetrical on the surface in the maxillary region and in five patients in the mandibular region.

Relationship between horizontal and vertical measurements of the neurocranium and face. Shortness of the horizontal measurement of the skull base width (t-t), of the width of the face (zy-zy), or of both was always associated with short maxillary (t-sn-t) and mandibular arcs (t-gn-t). Abnormal shortening of the horizontal measurements from the skull to the mandible were found in increasing frequency in these patients.

The vertical shortness of the lower third of the face (sto-gn) did not cause a decrease in the total height of the face (n-gn), but half of such patients had short maxillary arcs (t-sn-t), mandibular arcs (t-gn-t), or both.

Orbits

Abnormal measurements. The interocular diameter (en-en) was long (with a maximum of 41 mm) in 19 patients. The level of the palpebral fissures was unequal (with

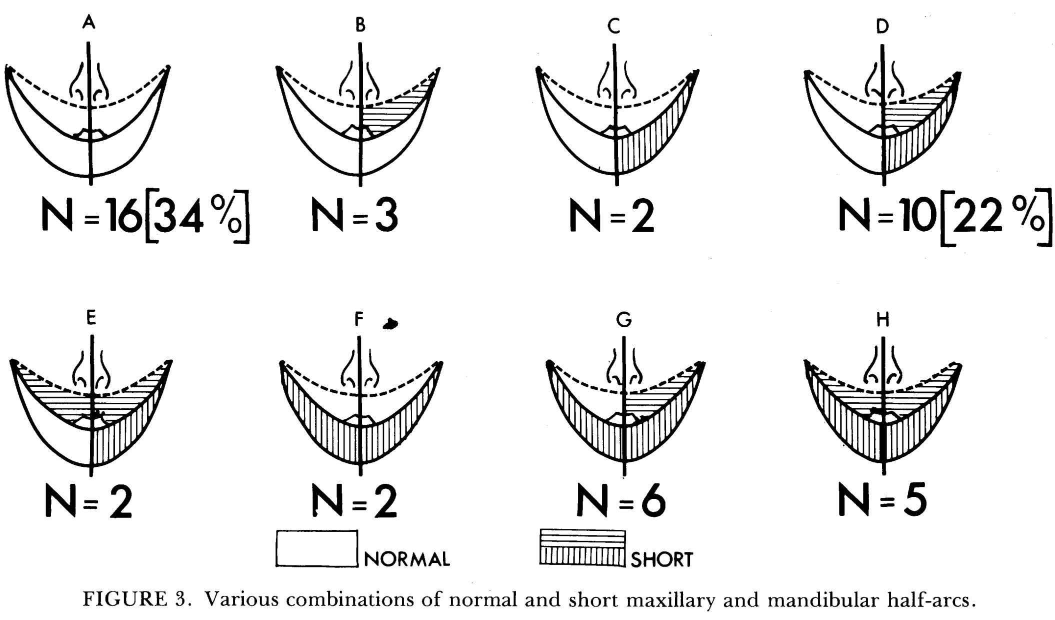

FIGURE 3. Various combinations of normal and short maxillary and mandibular half-arcs.

maximum difference of 3 mm) in 13 patients, and the biocular diameter (ex-ex) was short in one patient and long in another, the latter having a long interocular diameter (Figure 2).

Asymmetries. In 11 patients, the difference between the right and left endocanthion to midline distance was 2 to 3 mm. In 10 patients, the palpebral fissure lengths differed by 2 to 3 mm and were associated with some of the defects listed below.

Epicanthi were found in 14 of 44 patients (or 31.8%) over six years of age and in only 12 of 100 (or 12%) of the controls. One third of the patients with wide interocular diameter had epicanthus. Mongoloid eye fissure slants were found in six patients. Three patients had unilateral ptosis congenitalis and one of the three also had coloboma.

At least one of the above listed quantitative and/or qualitiative defects were seen in 31 of 46 patients.

Nose

Abnormal Measurements. The nose (n-sn) was long in 10 patients and the nasal bridge, the columella, or both were deviated 5° to 15° in seven. The nose was narrow (al-al) in three patients (Figure 4).

Patients with long noses always had elongated upper faces. The nasal bridge or columella or both were deviated to the side of the greater facial dysplasis in patients with moderate (5 to 9 mm) or marked (10 or more) asymmetry of the maxillary half-arcs. A short bi-alar diameter (al-al) was invariably associated with uni- or bilaterally short maxillary half-arc.

The most damaged part of the soft nose was the ala with 16 unilateral ala hypoplasias. The ala was flat or kinked in eight patients. A difference in columella lengths was noted in 11 patients (maximum 3 mm). In 25 patients, the nasal floor widths were asymmetrical (maximum 3 mm). The alar base (sbal) was more frequently dislocated in the horizontal plane (27 patients) with a maximum of 4 mm than in the sagittal direction (six patients). The deeper lying alar base was always associated with an ipsilateral short maxilla. Nostril asymmetry in 18 patients resulted from some of the above defects.

At least one of these quantitative and/or qualitative signs was seen in 37 out of 46 patients.

Upper Lip and Mouth

Abnormal measurements. The medial vertical upper lip length (sn-sto) was long in three patients, and the labial fissure length (ch-ch) was short in one (Figure 4). The vertically long upper lip was found in patients with elongated upper faces. The short labial fissure was combined with a markedly short maxillary arc.

Asymmetries. Asymmetry to a maximum of 4 mm between the lateral vertical heights of the upper lip (sbal-ls), a frequent sign (30 patients), was influenced by various factors (nasal ala base dislocation, nasal ala hypoplasia, vermilion irregularities, oblique slanting of the labial fissure) either singly or in combination.

The halves of the labial fissure without

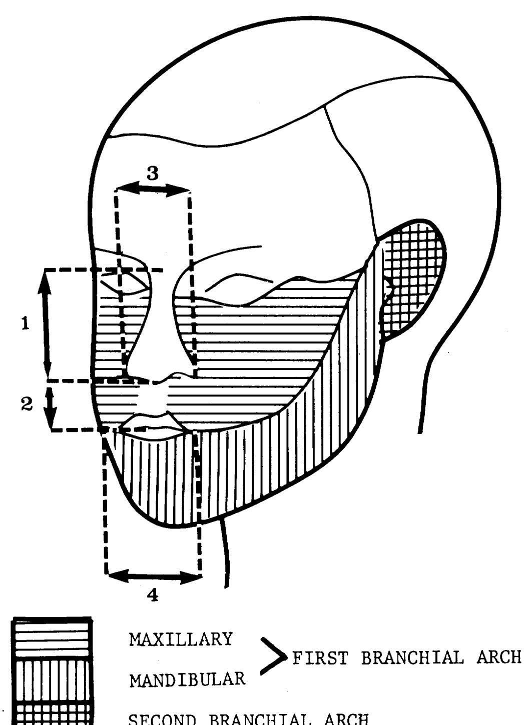

FIGURE 4. Measurements of the face schematically. 1 = length of the nose (n-sn = nasion-subnasale). 2 = medial vertical upper lip length (sn-sto = subnasale-stomion). 3 = width of the nose (al-al = alare-alare). 4 = length of the labial fissure (ch-ch = cheilion-cheilion).

any signs of lateral clefts were asymmetrical by 3 to 5 mm in four patients.

Three patients had unilateral macrostomia (commissural clefts), all with normal labial fissure length after surgery, one patient with a 7 mm difference between the labial fissure halves. The macrostomia located on the side of the facial damage was combined with ear aplasia, ear hypoplasia, or a low-set, normal sized ear, all with preauricular tags and fistulae. The nasal alar base was dislocated, and the ala was hypoplastic on the side of macrostomia.

At least one of the above listed metric and/or qualitative defects was found in 31 out of 46 patients.

EAR

Thirty-nine patients had microtic ears (27 aplastic and 12 hypoplastic). Six of these were low-set, and 20 had pre-auricular appendages, five with fistulae.

In seven patients with unilateral LFD, both ears were of normal size. However, on the side of the greater facial damage, the ear was lowset with asymmetrical inclination of the longitudinal axis (six patients) or had preauricular tags (four patients) or shape disfigurement (seven patients).

The radiological examination of the damaged side revealed narrowed auditory meatuses (two patients) and temporal bone deficiencies (five patients) in the patients with bilaterally normal sized ears.

Half of the normal sized ears combined with microtia also exhibited defects of location, inclination or, anterior surface design.

No patient was found with completely normal auricles.

Microtia and the location of the facial defects. Defects found exclusively on the side of the ear damage were dislocation of the endocanthion, deformity of the nasal ala, depression of the alar base, and macrostomia. The majority of cases showed, on the side of the microtia, a shorter maxillary or mandibular half-arc or both, shorter palpebral fissure, shorter columella and shorter lateral vertical upper lip length, longer endocanthion-facial midline distance, and wider nasal floor than on the less damaged side.

Degree of microtia and the shortness of mandibular arc. Both short (22) and normal (17) mandibular half-arc measurements were found on the side of the microtic ear. On the other hand, a normal sized auricle was seen on the side of the short (five) or normal half-arc (two).

A close relationship between the degree of microtia and abnormal mandibular measurements was observed only in those patients whose mandibular arcs were bilaterally short and differed from each other by 10 mm or more. Six out of seven such cases had an auricular aplasia and one ear was hypoplastic. The incidence of microtic ears markedly decreased as the degree of asymmetry between the two short mandibular half-arcs lessened.

Location of the associated facial defects. The defects observed on both sides of the face were coloboma, ptosis of the upper eyelid, mongoloid palpebral fissure slant, and epicanthus.

The horizontal alar base dislocation and nasal ala hypoplasia were found more often on the defective than on the other side of the face. The mouth commissure was often higher (14 patients) on the side of the facial defect than on the less affected side. In only one-third of these cases did the higher location indicate asymmetrical elongation of the labial fissure towards the damaged side of the face.

Discussion and Conclusions

Although the techniques of anthropometry (measurements) and anthroposcopy (inspection) are relatively simple, they "cannot give reliable results unless the observer has practised the methods thoroughly" (Weiner and Lourie, 1969). The criteria for the inspection techniques are liberally defined. Therefore, the number of signs assessed by inspection should be restricted to the minimum. Errors made at measurement may be directly related to the inexperience of the investigator.

The correct location of landmarks on the surface of the face and skull belong to the routine knowledge of anthropometry. However, in the deformed face (congenitally or traumatically), the surface landmarks may be dislocated or missing. In LFD patients, the sometimes missing "tragion" point, located on the tragus, was replaced by the point at the temporomandibular joint.

There was a wide spectrum of auricular defects ranging from ears of normal size (15%) but of abnormal location to inclinaton and shape disfigurements and various types of microtia (85%).

Generally, the relationship between the extent of the facial damage and the degree of microtia was not apparent. However, a severely deficient mandibular arc was usually associated with an aplastic ear.

Although asymmetry in the maxillary and mandibular region was a frequent sign, there were a few patients who exhibited either symmetrical or very slightly asymmetrical half-arcs. It was an unexpected finding that, in this unilateral syndrome, almost one-third of the study group had bilaterally short maxillary or mandibular half-arcs or both.

Because the bitragion-menton arc covers a wider area than the mandible, the mandibular surface arc does not reflect all skeletal defects, only those of great extent.

The preauricular tag, always found on the damaged side of the face when located in front of a normally sized auricle, was an indicator of deviant location and defective temporal bone structures. The preauricular appendage may well be a minor manifestation of the disturbed ear development (Converse el al., 1973; Pruzansky, 1973).

Frequent defects in orbits, nose, upper lip, and mouth were observed on both sides of the face, with greater concentration on the side of the mandibular or auricular damage.

In severe cases of the syndrome, the face showed narrowing in horizontal facial diameters and arcs and prolongation in the vertical profile measurements of the upper face.

Anthropometry used in assessment of congenital anomalies of the face is relatively new. Further studies will necessitate modification of the classical techniques or development of new measuring methods.

Both x-ray cephalometry and surface (facial) cephalometry have their limitations. A combination of both methods seems to be a logical solution to the problem.

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