10 Facial Growth and Morphology in the Unoperated Cleft Lip and Palate Subject: The Sri Lanka Study

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Studies of subjects with unoperated clefts of the lip and palate have been undertaken for the past 80 years [1–29].

Adults with unoperated clefts of the lip and palate provide the ideal control group for investigators studying the natural history of facial growth and morphology in these subjects. The absence of surgical intervention provides an opportunity to study the outcome of facial growth, morphology, and speech, using an absolute comparative baseline. An evaluation of the intrinsic versus the potential iatrogenic influences can be separately analyzed, when this group is compared to conventionally operated subjects. They also highlight the discrepancies in health care provision between the wealthy western world and the developing/least developed world.

Withdrawal of surgery for research purposes would be unethical where such facilities are available. Likewise, assignment to surgical or nonsurgical management programs on a prospective random allocation basis could not be permitted. For these reasons, studies on the unoperated subject have been made on individuals from the developing world, where surgery may not be readily available. This has led to many of the limitations listed below being evident in such studies. Additional limitations have been imposed by the authors themselves, using inappropriate management of the material collected.

The difficulties experienced by research workers in the field of cleft lip and palate are seen in the limitations of many studies. These may be summarized as follows:

1. Small sample size
2. Wide age distribution
3. A narrow age distribution of very young subjects
4. Mixtures of unoperated, partially operated, late operated and early operated subjects
5. Mixtures of subjects from different cleft types
6. Males and females grouped together
7. No controls from the normal population
8. Controls of treated patients from the same population seldom available (applicable to studies of unoperated versus operated subjects)
9. Few postoperative follow-up studies where late surgery has been performed

Unquestionably, the major problem is that of small sample size, which has been responsible for the inappropriate handling of the available data.

The general consensus in the literature relating to the unoperated UCLP subject suggests that they have the potential for near normal facial growth. However, serious limitations are obvious in the analysis of the data, especially by attempts to increase the sample size by pooling nonhomogeneous groups. Despite limitations of all previous studies into facial growth in the totally unoperated cleft lip and palate subject, the results suggest that facial growth proceeds reasonably well. Midface depth, the parameter of most concern in CLP, is not compromised in unoperated subjects.

10.1 Sri Lankan Cleft Lip and Palate Project

The Sri Lankan Cleft Lip and Palate Project has developed as the largest multidisciplinary surgical and research program concerned with the unoperated cleft lip and palate subject [30]. Since 1984, extensive records have been collected on over 1,000 subjects, on whom 820 operations have been performed. There have been 13 visits from 1984 to 2002. The aims have always been threefold: treatment, teaching, and research based on a multidisciplinary team. Over 50 professionals have been involved, many having attended on over 10 visits.

The unresolved conflicts of opinion regarding the aetiology of facial growth distortion in repaired cleft lip and palate subjects were the main reasons for the establishment of this Project. The possibility of

Table 10.1. Unoperated groups

	UCLP				BCLP				ICP			
	Young		Mature		Young		Mature		Young		Mature	
	M	F	M	F	M	F	M	F	M	F	M	F
Sample size	15	8	21	11	5	6	5	8	8	15	9	9
Age range	13–20	14–18	21–47	19–43	15–19	14–19	21–49	21–55	13–20	13–18	21–44	19–43
Mean age	15.5	15.8	28	26.8	17.2	16.6	30.2	32	N/k	N/k	N/k	N/k

Table 10.2. Control group

	Young		Mature	
	Male	Female	Male	Female
Sample size	21	58	24	16
Age range	14–19	14–18	20–30	20–30
Mean age	15.9	16.2	25.6	27.5

addressing these questions by studying "nature's experiment" on hundreds of unoperated and late-operated subjects of all ages from birth to old age was unique. Such an opportunity is rare. Investigators usually have to resort to animal experiments in order to provide sufficient numbers in a controlled manner [31, 32].

This chapter, which deals with totally unoperated subjects from Sri Lanka presents a descriptive account of facial morphology, a cephalometric analysis (UCLP, BCLP, and ICP groups), a GOSLON yardstick analysis (UCLP group), and a reflex microscopic analysis of dental study models (UCLP and ICP groups). The material comprises 55 UCLP, 23 BCLP, and 41 ICP subjects (Table 10.1), and 119 healthy control Sri Lankan subjects (Table 10.2). All unoperated subjects described in this chapter were over 13 years of age when they first presented.

Because the onset of puberty is some two to three years later in Sri Lanka than in the West, it was necessary to separate pre- and postpubertal groups [33]. Further, there are significant differences in the timing of the onset and termination of puberty between males and females. This, and the fact that there are some significant differences in facial morphology between the sexes, has necessitated their separate analysis.

10.1.1. Controls

One hundred nineteen healthy adult Sri Lankan non-cleft subjects provided Control data. These comprised 40 male medical students and 79 female hospital nurses and medical students. The Controls were aged between 20 and 30 years of age (Figs. 10.1, 10.2).

10.1.2 Records Collected for Study

Lateral skull radiographs and dental study models of the unoperated subjects comprise the material for cephalometric and Goslon Yardstick analyses.

10.1.3 Radiographs

The lateral skull radiographs were taken in a cephalostat sited in a private sector hospital in Galle. The same design of cephalostat was used in Kandy and Galle on all subsequent visits.

In 1984, the protocol for setting up the radiographic apparatus was established and supervised for each exposure by the author. In subsequent visits, all orthodontic members of the Team, after training, took turns in placing the subjects in the cephalostat and supervising all exposures.

Great care was taken at each radiographic session to ensure that the anode to midsagittal distance was precisely 152.5 cm (the Imperial measurement of 5 ft) and the midsagittal plane to film distance was 16 cm. The central ray was arranged at right angles to the sagittal plane. This was determined by an electric light source within the anode housing, casting a superimposed shadow of both ear rods on a sheet of white paper which was attached to the x-ray film cassette. These measures ensured reproducible skull radiographs on all occasions with consistent magnification error. The patients were posed with the teeth lightly occluded in maximal intercuspation and the Frankfort plane parallel to the ground.

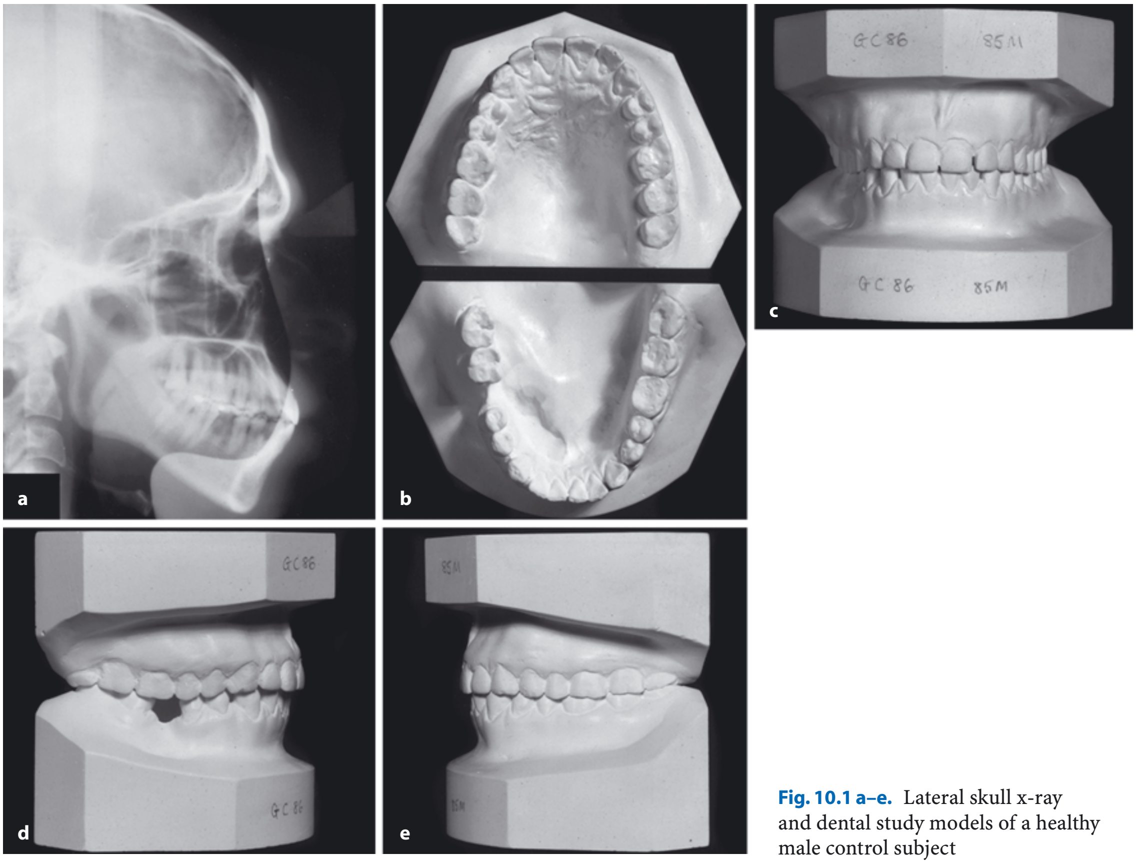

Fig. 10.1 a–e. Lateral skull x-ray and dental study models of a healthy male control subject

Table 10.3. Mature unoperated subjects cephalometric results (UCLP, BCLP, and ICP)

	Controls Mature		UCLP Mature		BCLP Mature		ICP Mature	
	M	F	M	F	M	F	M	F
Ba_N Cranial Base	112.5±2	108±2	110±2	110±4	102.3±7	100.2±8	108.4±5	100.3±5
SNA Maxillary protrusion	85.4±3	85±3	85±2	86±4	90.3±10	90.6±8	83.2±6	83.4±4
SNANS Basal maxillary protrusion	87.1±3	87.1±3	84±2	86±4	94.6±10	94.5±8	84.7±6	85.3±5
Ans-Ptm Palatal Length	57.8±5	53.1±4	54±2	51±2	61.6±6	61.3±6	50.3±6	49.2±5
SNB Mandible protrusion	82±4	83.4±3	80±2	81±3	81.6±5	81.7±6	81.3±5	81.3±4
AR-TGO Ramus height	57.5	50.0	51.3	48.9	48.5±4	47.8±41	48.6	47.1
S_N_ANS Upper face height	54.5	51.5	52.6	47.5	46.6±3	45.5±2	49.2	45.6
N-S-Ptm Upper post face height	44.0	40.3	38.3	37.0	33.5±4	31.2±3	39.7	33.4

Temperatures were always around 30 °C, and the humidity was often near 100%, but this did not adversely affect the quality of the films, which were immediately processed in wet tanks. They were dried and suspended from wire "washing lines" strung across the main x-ray room. Patients were detained until after their x-ray had been examined, in case a repeat x-ray was required (Fig. 10.3, Table 3).

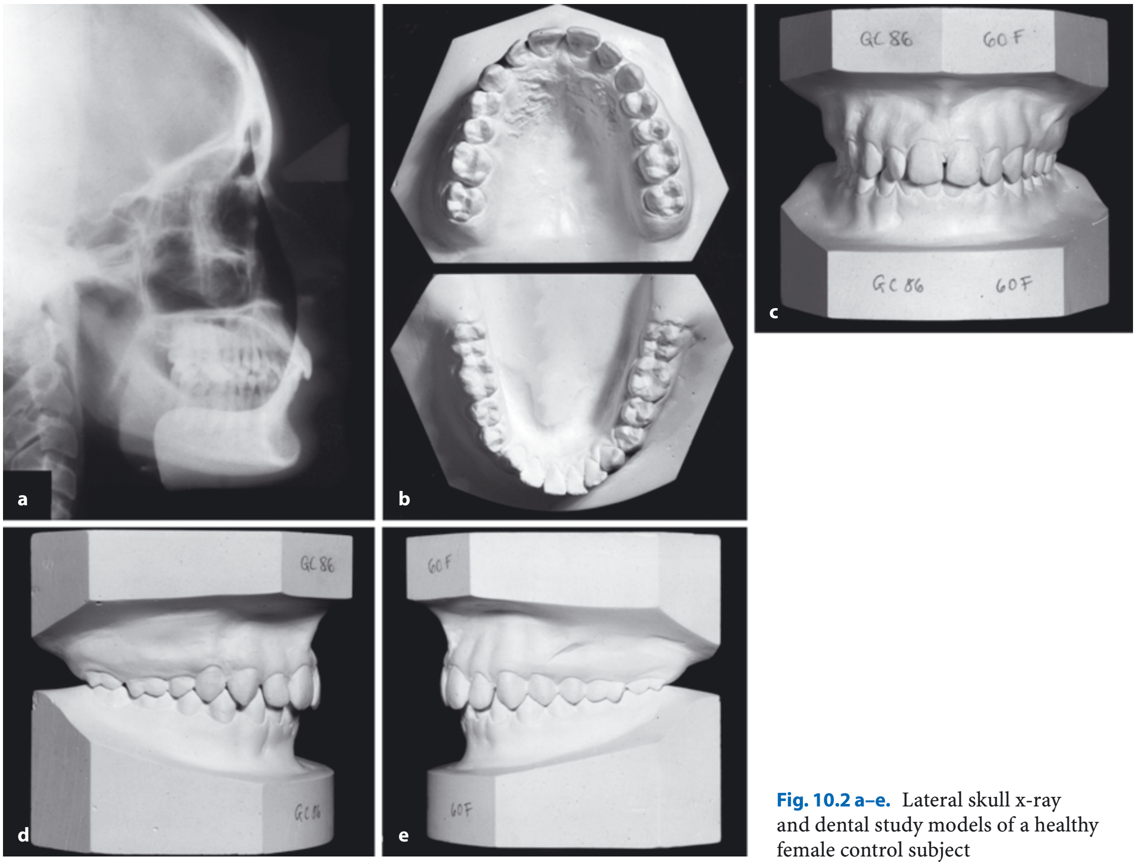

Fig. 10.2 a–e. Lateral skull x-ray and dental study models of a healthy female control subject

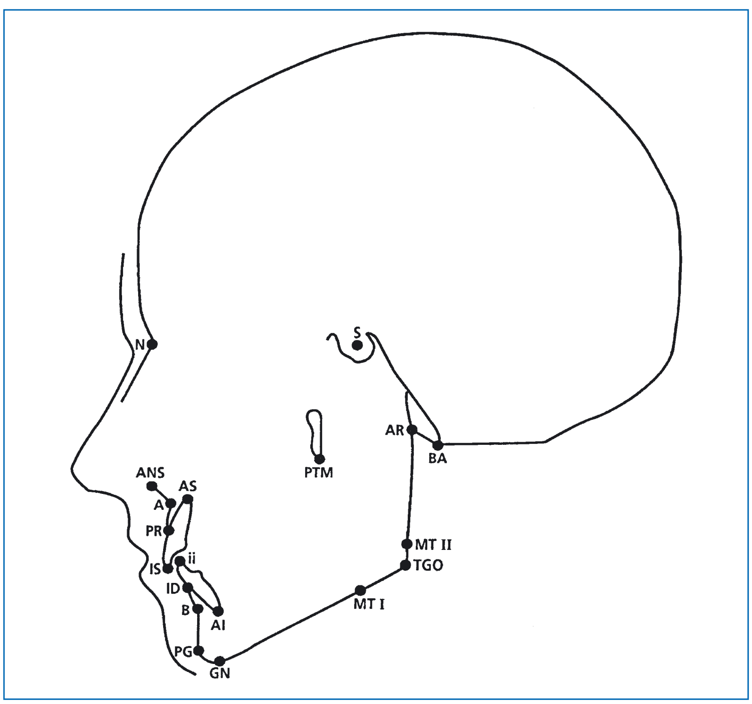

Fig. 10.3. Digitized points used for cephalometric analysis

10.2 Unoperated Unilateral Cleft Lip and Palate

The most striking feature in the unoperated unilateral cleft lip and palate subject is the protrusion of the upper labial segment. Subjects present with large overjets, proclined upper incisors, eversion of the major segment and a mild contraction of the lesser segment in the anterior region. Buccal cross-bites are rare. Cephalometric analysis displays normal cranial base measurements and protrusion of the maxilla relative to the mandible. No cases presented with maxillary retrusion (a common feature in the operated case). Figures 10.4 and 10.5 illustrate typical examples of the facial appearance and dental study models of unoperated unilateral cleft lip and palate cases.

10.2.1 Dental Study Models

Impressions were taken in alginate material with the patients sitting upright on a wooden chair. Bite registration was made using conventional denture wax. In 1984, the author and accompanying oral surgeon cast the impressions in white dental plaster in the hotel bathroom after the day's work. This process was fraught with difficulty because dental plaster sets extremely rapidly in the tropics.

On all subsequent visits, a local Sri Lankan technician was employed. Impressions were cast in dental stone on the day that they were taken, incorporating the tooth bearing and important anatomical areas. Care was taken to include all anatomically visible structures. This necessitated modifying aluminium

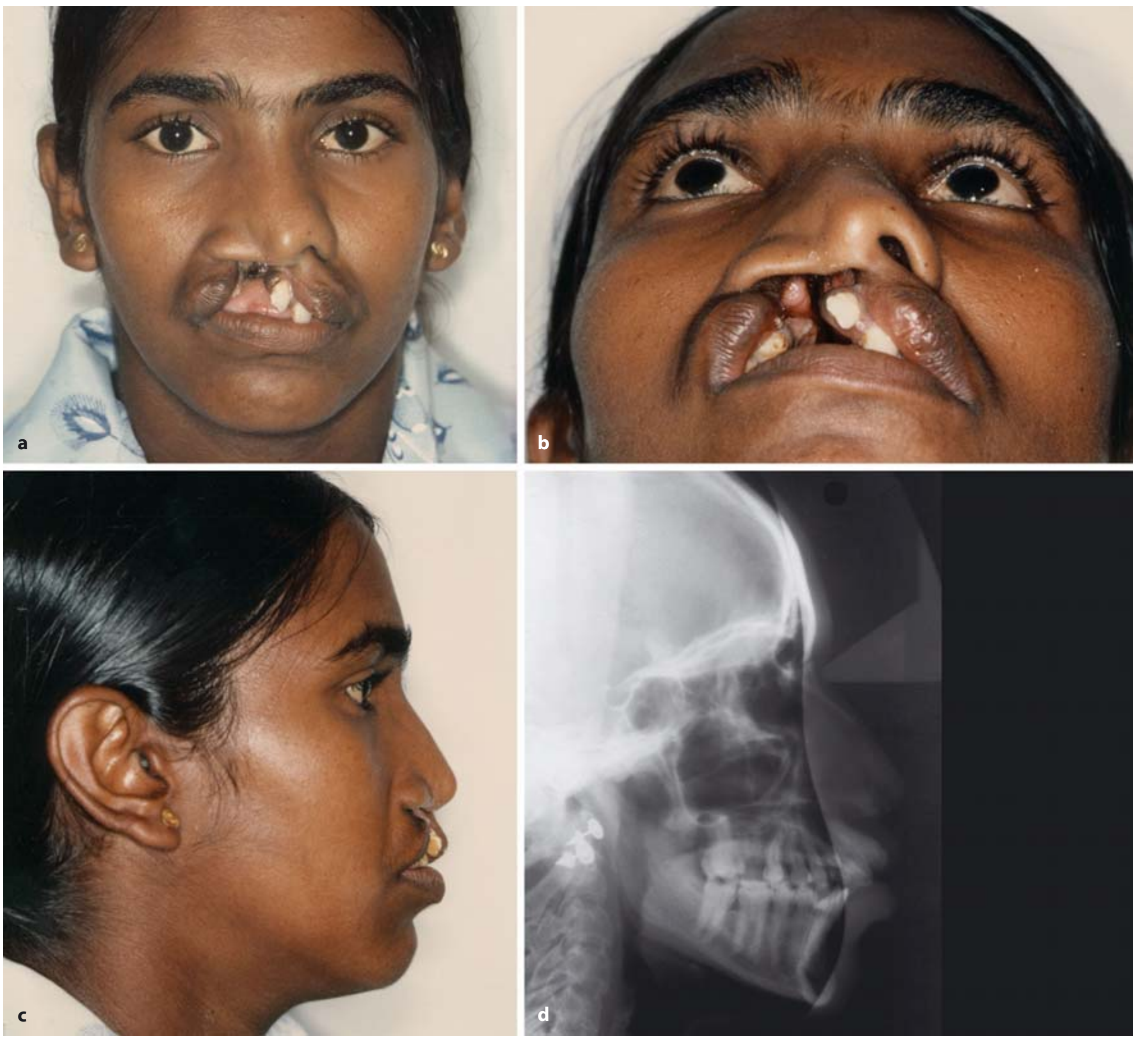

Fig. 10.4 a–j. Adult female subject with complete unilateral cleft of the lip and palate

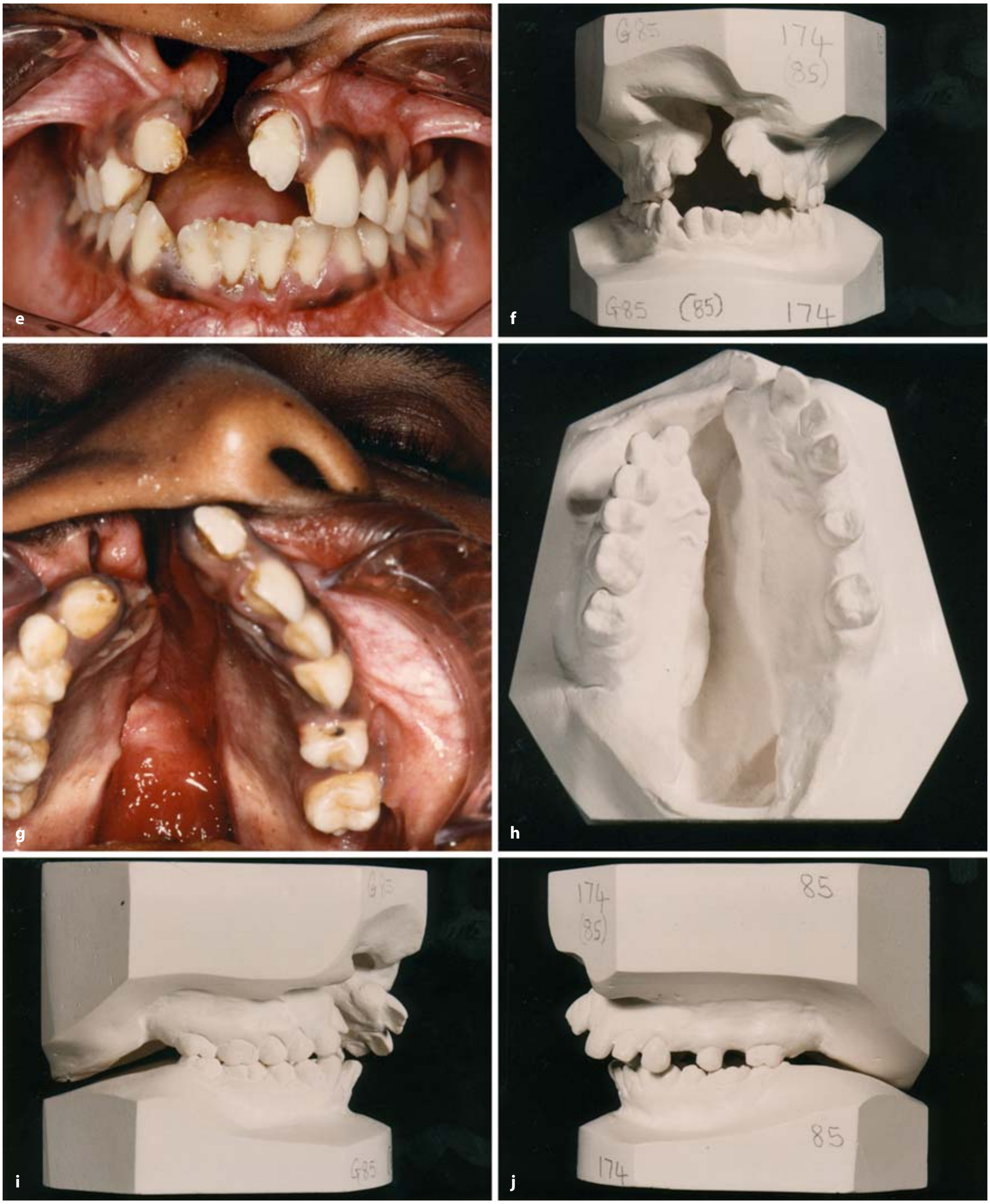

Fig. 10.4 a–j. *(continued)*

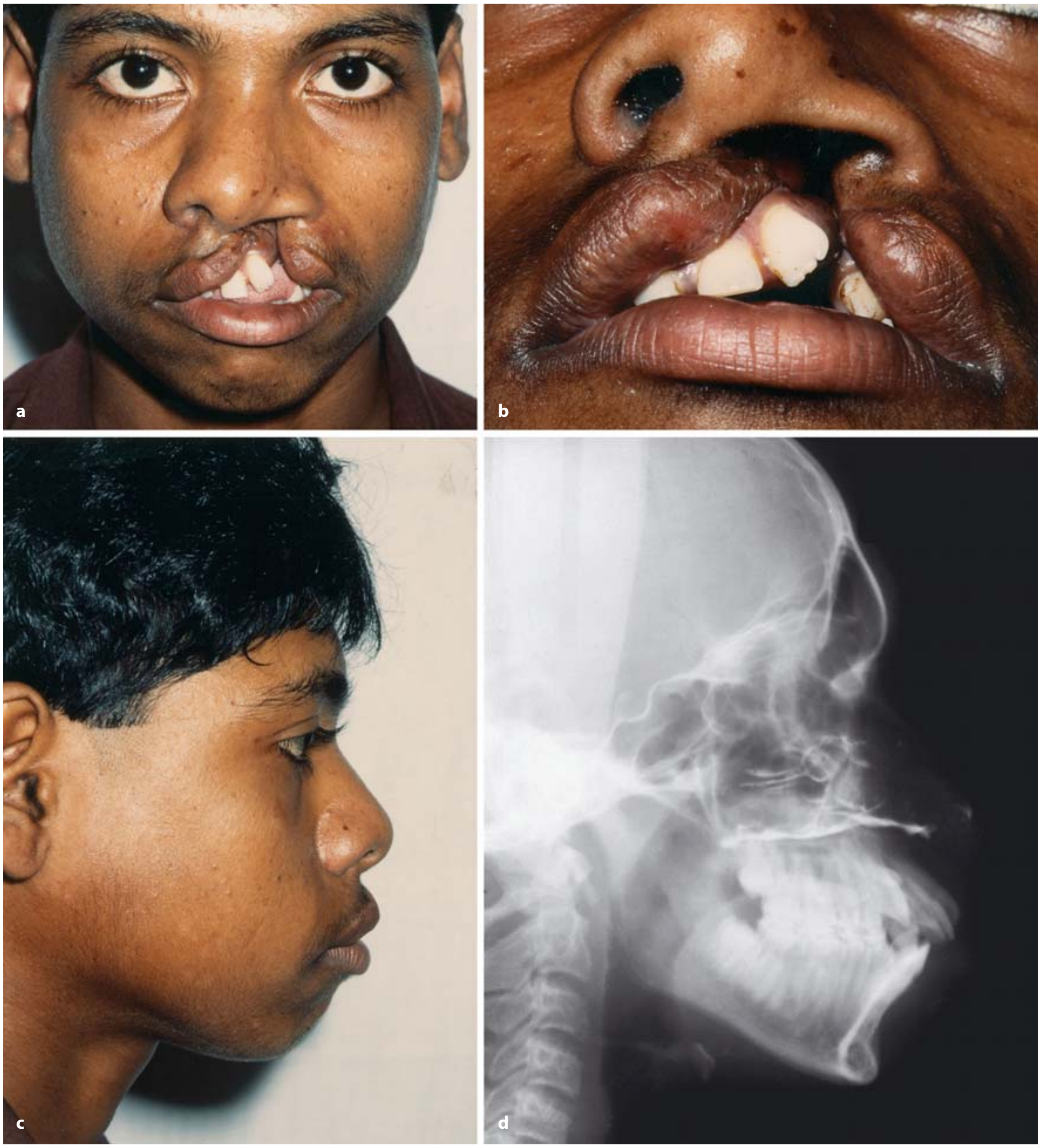

Fig. 10.5 a–j. Adult male subject with complete unilateral cleft of the lip

trays with white wax rims. Huge volumes of alginate were used on some of the adults with wide-open clefts, and included impressions of the middle and inferior turbinate bones, the entrance to eustachian tubes, and the posterior wall of the pharynx. Some impressions utilized 13 scoops of alginate powder. None of the patients vomited. Figure 10.6 shows alginate impressions with extension beyond the inferior turbinates, the entrance to the eustachian tubes, and the posterior wall of the pharynx.

Final orthodontic trimming and basing of the models was performed at the laboratory in the Maxillofacial and Dental Department in the Hospital for Sick Children, Great Ormond Street, London.

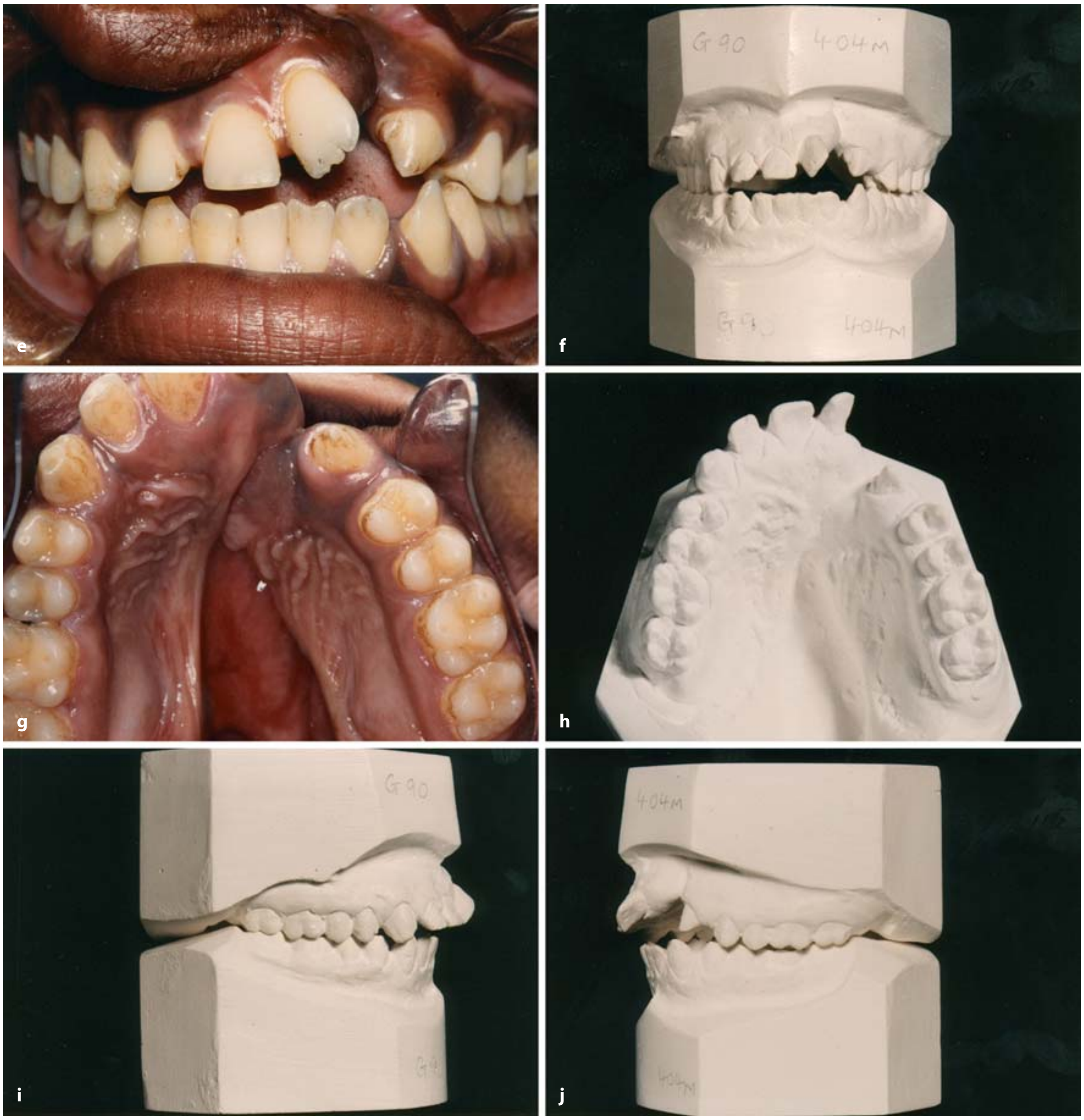

Fig. 10.5 a–j. *(continued)*

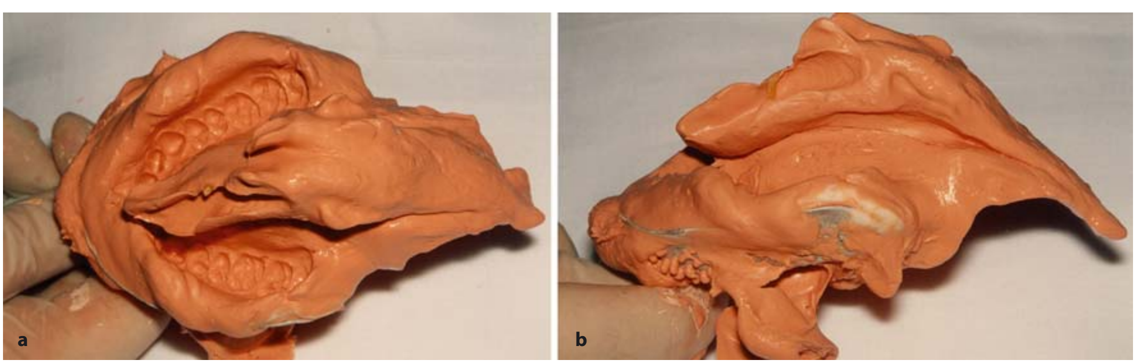

Fig. 10.6 a, b. Upper arch alginate impressions, maximally extended beyond the inferior turbinates, the entrance to the eustachian tube and the posterior wall of the pharynx

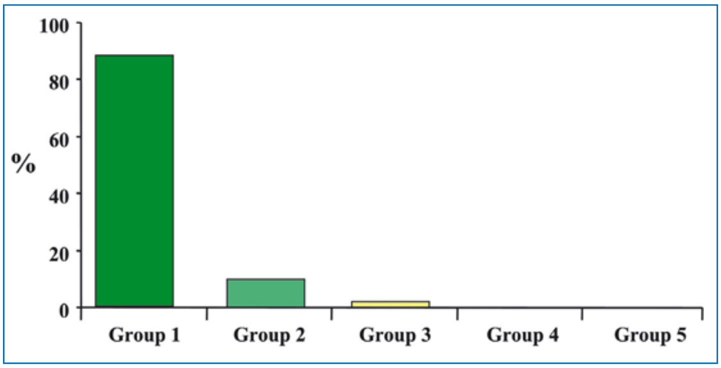

Fig. 10.7. GOSLON Grouping. Sri Lankan Unoperated UCLP. n = 51; 98% in Excellent (1) or Good (2) arch relationships

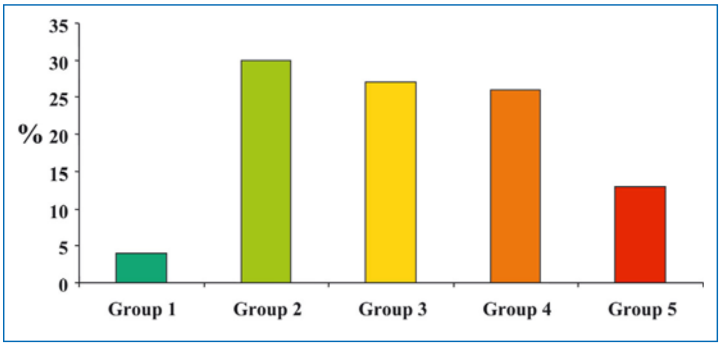

Fig. 10.8. UK CSAG Study 1998. GOSLON CSAG (Clinical Standards Advisory Group) GOSLON results of the whole of the UK

10.2.2 The GOSLON Yardstick

The GOSLON Yardstick [34] a London/Oslo consensus of 5 groups of increasing deformity in dental arch relationships was applied to 49 unoperated cases for whom cephalometric analysis had been performed. Two additional cases where study models, but not lateral skull radiographs, were available have been included.

"UNOPS" in GOSLON STUDY(UCLP) [51]

Younger:	Male	Female
	13	8
Mature:	Male	Female
	19	11

The ranking was performed by the author. Previous reports have demonstrated the reproducibility of the author's assessments, and the robustness of the Yardstick in the discrimination between differences in samples from different centres [35, 36]

The GOSLON yardstick results showed 98% of the cases in group 1 or 2 (excellent or very good arch relationships) and no cases in groups 4 or 5 (Fig. 10.7).

Because the yardstick is not a direct measure of skeletal morphology or growth, it is possible to pool males and females over 13 years in a single group. These results are in marked contrast to those of any centre worldwide examining operated patients, where only a very small minority of patients are found in group 1 (Fig. 10.8).

10.2.3 Unilateral Cleft Lip and Palate Study Models Analysis by Reflex Microscope (Fig. 10.9)

10.2.3.1 Arch Widths

The cleft group was narrower in the second molar region by a mean of 1.6 mm and narrowed progressively to a maximum mean deficit of 5.0 mm in the canine region, resulting in the arches being more V shaped. There were no differences according to sex.

10.2.3.2 Tooth Widths

There were significant differences in tooth size between the cleft group and the controls: the teeth of the cleft group were consistently smaller and the largest differences were in the central incisors. No significant differences were found in comparisons of the cleft and noncleft sides, sexes, or side to side within each group.

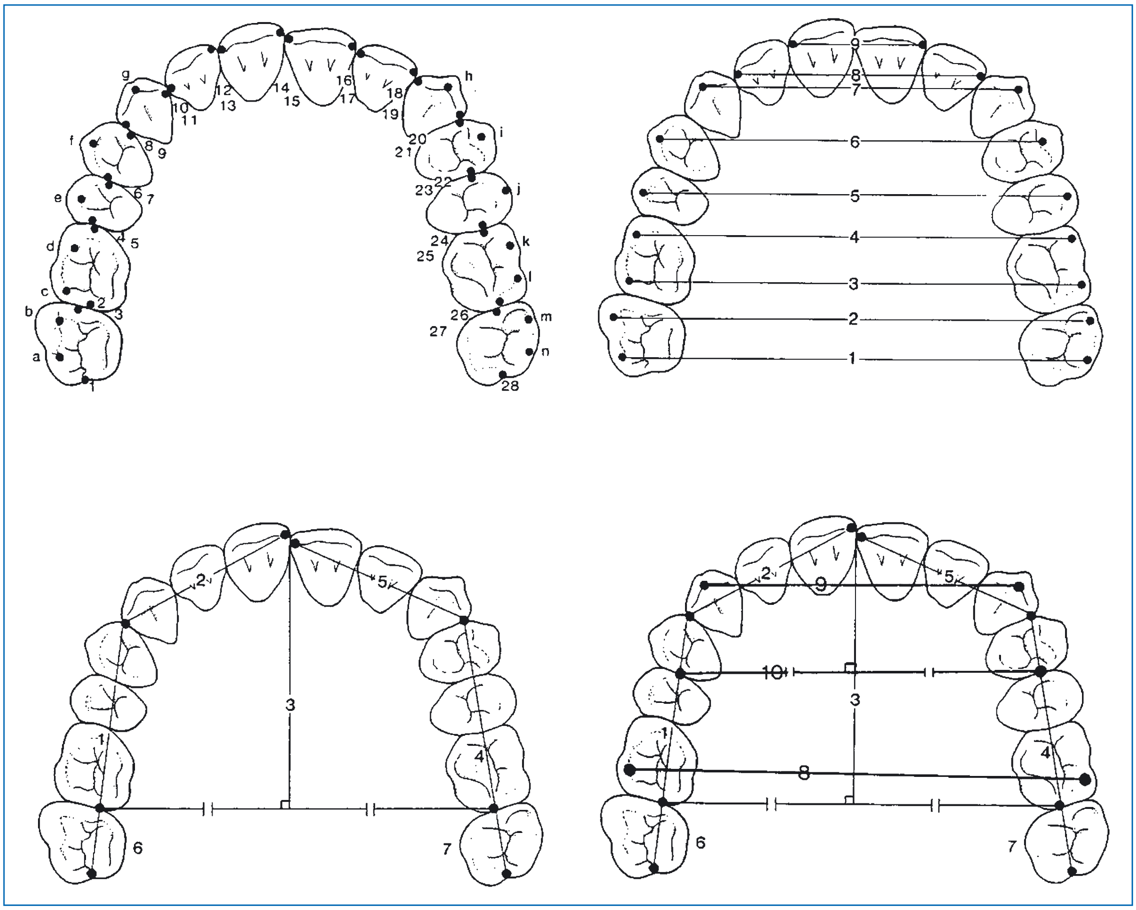

Fig. 10.9. The digitized points, chord lengths, and arch widths used in the reflex microscopic analysis of study model

10.2.3.3 Chord Lengths

No significant differences were found in chord lengths.

10.2.3.4 Crossbites

There was a higher prevalence of crossbites in the cleft subjects (8 cases) than in the control group where there were none.

10.2.3.5 Overjet

The overjet was much larger in the cleft group. The mean overjets in the cleft group was 8.2 mm compared to 3.7 mm in the controls

10.2.3.6 Missing Teeth

The percentage of missing teeth was higher in the cleft group; the most commonly missing teeth in the cleft group were the maxillary lateral incisors.

10.2.3.7 Crowding

There was no crowding in the buccal segments of either the control group or the cleft group. In fact, all dental arches examined were very well aligned and some spacing was generally evident.

10.2.4 Summary of Reflex Microscope Findings on Study Models: UCLP [15]

Tooth size and arch widths that were found to be smaller in the unilateral cleft lip and palate subjects than in the control group would suggest that there is some degree of primary hypoplasia in this group. However, these differences are small and would not account for the gross maxillary retrusion frequently reported in surgically repaired unilateral cleft patients. Even with this small primary hypoplasia, overjets in the cleft group are larger than the controls and the prevalence of crossbites is surprisingly low. These results concur with the cephalometric findings of a smaller retrusive mandible in the unilateral cleft and palate subject.

These features demonstrate the intrinsic potential for these subjects to grow relatively normally with minor distortions around the cleft site itself where the dentition is unrestrained because of the disrupted musculature. There are, however, some intrinsic growth deficiencies, particularly a shortened ramus height, and reduced upper anterior midface height and upper posterior face height.

10.3 Unoperated Bilateral Cleft Lip and Palate

Unoperated bilateral cleft lip and palate cases demonstrate massive protrusion of the premaxilla with gross proclination of the upper incisors. Interestingly, they show significantly smaller cranial base lengths. There are intrinsic growth deficiencies; a shortened ramus height, and reduced upper anterior midface height and upper posterior face height. The initial characteristics of the newborn bilateral cleft lip and palate subject persist during growth. The prominent premaxilla, wide alar bases, often laterally deviated premaxillary segment, short columella, and rudimentary prolabium with no muscle attachment are characteristic features. Figures 10.10–10.12 illustrate the typical examples of the facial appearance and dental study models of unoperated bilateral cleft lip and palate cases [37].

Fig. 10.10 a–d. Mature male BCLP, note lateral segments have moved to the midline excluding the premaxilla anteriorly

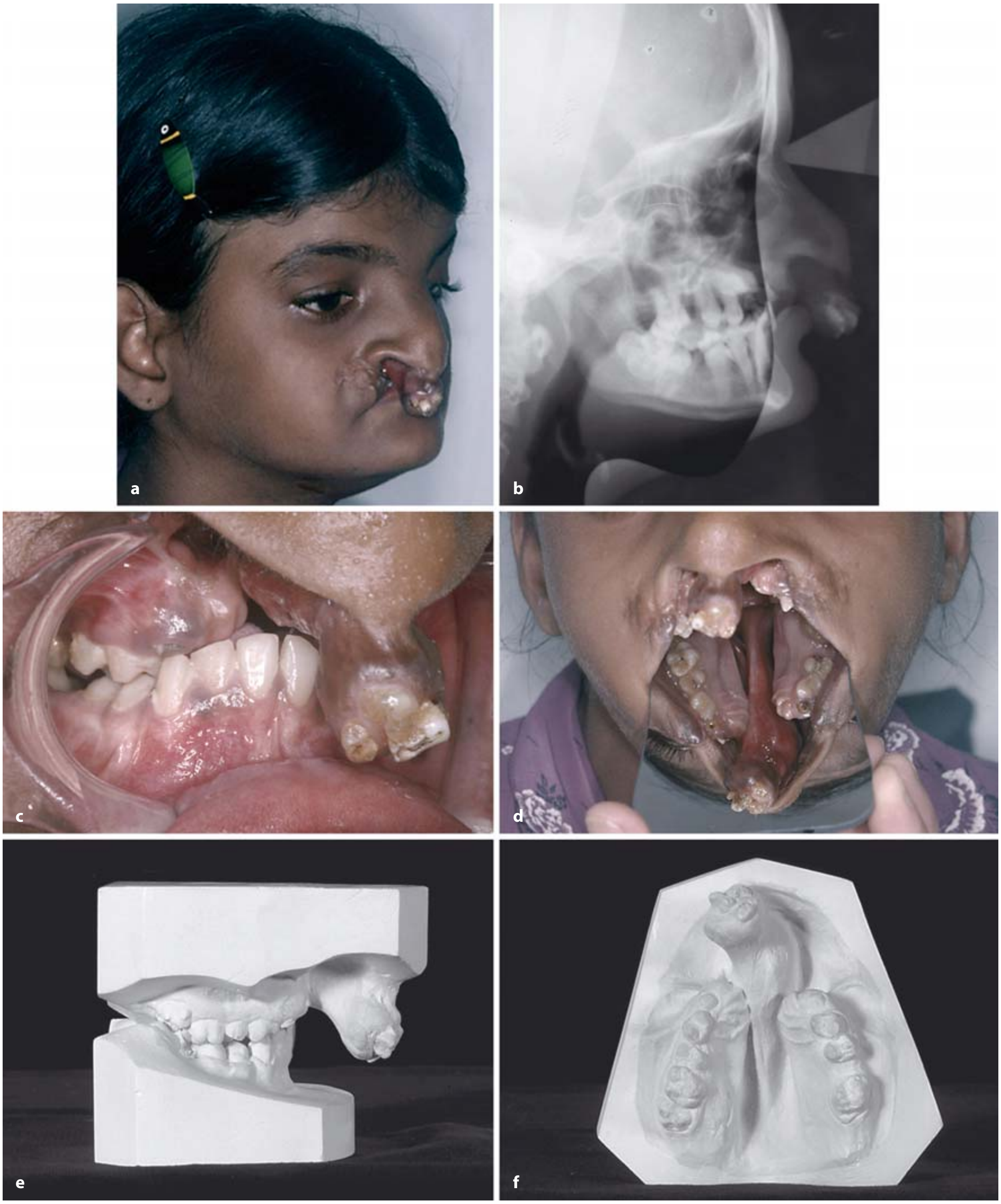

Fig. 10.11 a–f. Female unoperated BCLP unrestrained forward and downward growth of the premaxilla

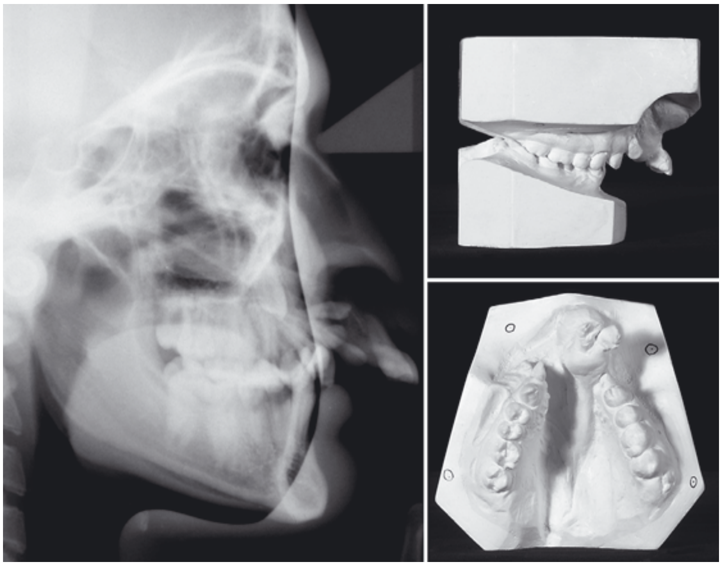

Fig. 10.12. Male lateral skull x-ray and study models of unoperated BCLP. Note gross unrestrained forward and downwards growth of the premaxilla

10.4 Unoperated Isolated Clefts of the Palate

Unoperated isolated clefts of the palate present with relatively normal appearance. However, this group demonstrates more intrinsic deficiencies than clefts of the lip and palate. They have normal upper and lower dental arch relationships but with bimaxillary retrusion. They present with short maxillary length, small mandibles, and reduced upper anterior and posterior face heights. Figures 10.13–10.15 illustrate typical examples of the facial appearance and dental study models of unoperated isolated clefts of the palate.

10.4.1 Isolated Cleft Palate Study Models Analysis by Reflex Microscope [38]

10.4.1.1 Tooth Sizes

The tooth sizes in clefts of the secondary palate were found in general to be smaller than in the normal group, although these differences were not large and were clinically unimportant. The greatest differences were found in the incisor region. No differences were found between the sexes of the same group.

10.4.1.2 Chord Lengths and Perpendicular Distances

Chord lengths from the first molar to canine region were not different between the cleft and the control group but were different in the canine to central incisors distance. This reflects the consistently smaller size of the teeth in the patients with clefts of the secondary palate. In addition, perpendicular distances were generally smaller.

10.4.1.3 Arch Widths

The arch widths of the clefts of the secondary palate were narrower than the controls. This may be an adaptation of the dentition to the smaller and narrower mandible in this group.

10.4.1.4 Overjets

The overjets in the cleft group were slightly increased compared to the control despite the fact that the chord lengths and the perpendicular distances were shorter. This again may be a reflection of small mandibular size.

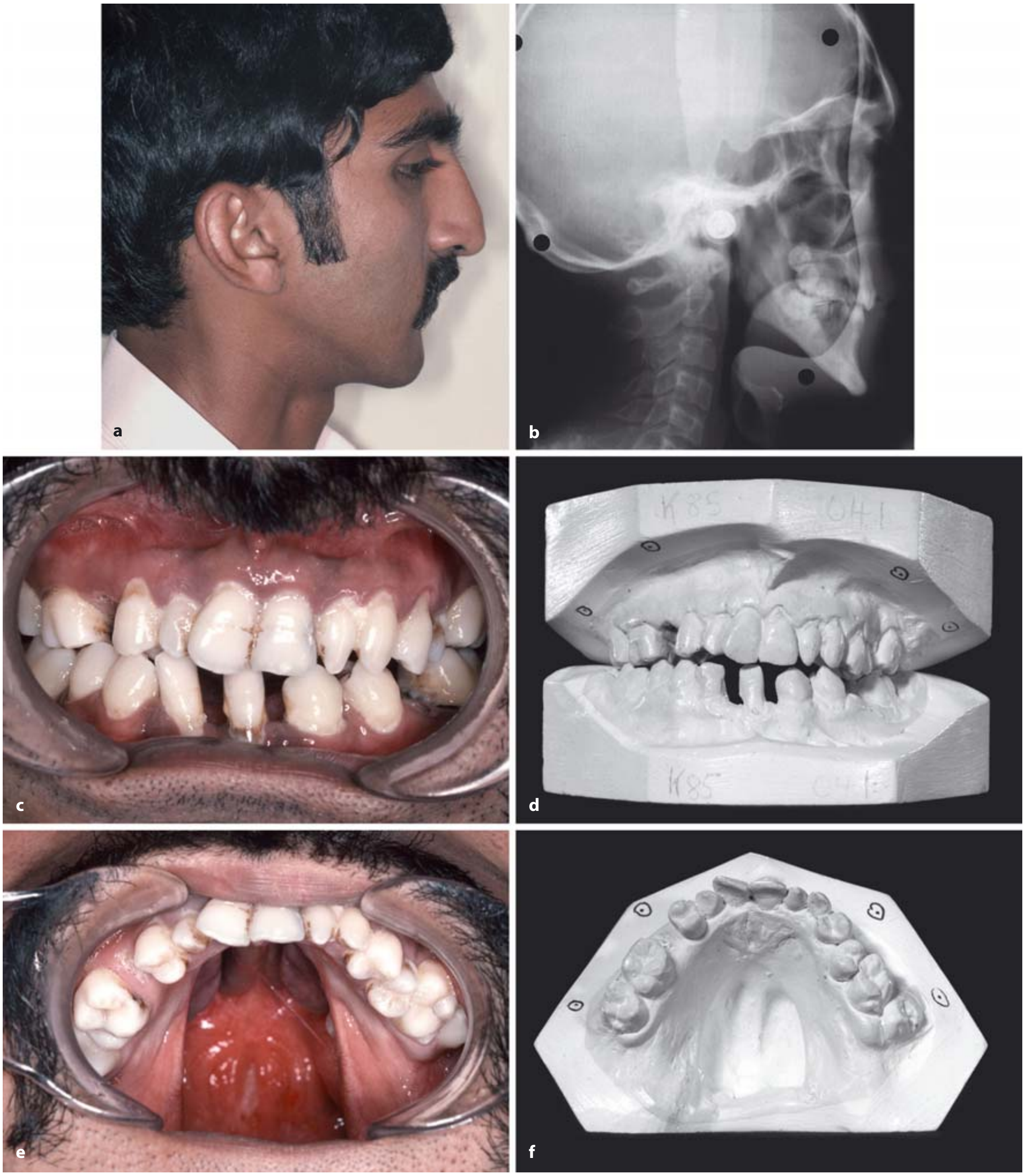

Fig. 10.13 a–f. Adult male subject with isolated cleft of the hard and soft palate

Fig. 10.14. Mature female with U-shaped ICP

Fig. 10.15. Mature female with V-shaped ICP

10.4.2 Factors Influencing Interpretation of Results from the Sri Lankan Cleft Lip and Palate Project

10.4.2.1 Malnutrition and Growth

It should be recognized that the above studies are derived from subjects in the developing world. Although Sri Lanka is a relatively advanced developing country, there is nevertheless significant malnutrition and endemic infections, for example, malaria.

The failure of infants with clefts to gain weight adequately has been documented by several authors [39, 40]. There is increasing evidence to suggest that poor nutrition in early life may be an important factor in growth disturbances seen in later life [41].

However, if there is failure to thrive or no catch-up growth by the age of 2 or in some papers by 5 years then perhaps attainment of normal limits for height, weight, or body mass index can never be expected.

Malnutrition, particularly at a period of especially rapid growth such as in utero, has long-lasting effects. Subsequent influences of undernutrition in the first and second years of life or later in childhood leave a long-lasting, complex growth problem. During childhood, stature is determined by the size that an infant has reached by the end of the first year of life, which is partly determined by genetic circumstances and influenced greatly by nutrition and the subsequent rate at which the child grows.

Nutritional status has a profound effect on growth hormone secretion. Malnutrition is a well-recognized form of reversible growth hormone resistance, which can be normalized with nutritional supplements. A malnourished mother is likely to give birth to a baby with low birth weight, while children with protein-energy malnutrition do not grow as well as others, according to a recent report [42]. This kind of malnutrition is an underlying cause of almost one third of the deaths among children under 5 years in Sri Lanka. Malnutrition is still a serious problem in Sri Lanka [43]. Food insecurity is one of the major reasons for malnutrition in Sri Lanka according to the Department of Census and Statistics. Poor financial and physical access to food is responsible for the malnutrition and food insecurity. Drastic price increases of essential food commodities and stagnating or deteriorating incomes created poor financial access to food. The civil war from 1984–2002 in Sri Lanka has exacerbated the essential food and financial problems.

A recent survey of 16,000 Sri Lankan children found that only one quarter were properly nourished [44]. More than one third were suffering from third degree malnutrition, the level beyond which children exhibit distended stomachs and skinny frames. Supporting evidence from the National Peace Council indicated that only 4,863 children under 5 years out of a random sample of 16,767 were within normal nutritional limits. Six thousand three hundred and seventy one children had third degree malnutrition, 3,186 with second degree malnutrition, and 2,347 with first degree malnutrition [45]. According to this report, diseases such as malaria, which is still prevalent in Sri Lanka are a primary cause of malnutrition. Worm infestations are the second most common cause. Lack of food itself is the third cause.

In another supporting paper it was found that Sri Lankans require a calculated average of 2,260 calories per day. Availability of protein has gradually increased; nevertheless a high incidence of malnutrition exists with 60% of children under 5 suffering from malnutrition [46, 47]. Poor growth of preschool children, high rates of low birth weight babies, poor maternal nutritional status, and micronutrient deficiencies are common nutritional problems in Sri Lanka.

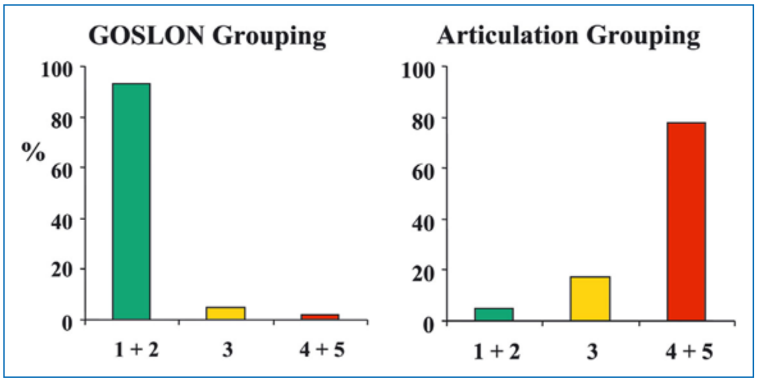

Fig. 10.16. Outcome for unoperated subjects beyond 13 years of age (n = 42). In both GOSLON and Articulation groupings 1 and 2 are excellent or good, while groups 4 and 5 are bad or awful

To measure quality of life in a nation, the United Nations Development Program [48] started figuring a Human Development Index (HDI). A nation's HDI is composed of life expectancy, adult literacy, and gross national product per capita.

There are vast differences when comparing or studying a different ethnic culture. The HDI for the UK is ranked as number 13 out of 130 nations. Sri Lanka is ranked at 79 – much lower than the UK. The comparisons between the two countries are shown in the table below.

UK and Sri Lanka Human Development Index

	UK	Sri Lanka
Life expectancy (years)	77.7	72.1
Total population (millions)	59.4	18.9
Annual population (growth rate)	0.1 %	0.8 %
Population under age 15	19 %	26.3 %
Undernourished people	0	23 %
Children underweight for age	0	33 %
Children underheight for age	0	17 %
Infants with low birth weight	8 %	17 %
Malaria cases (per 100,000 people)	0	1,111

Nutrition or subsequent malnutrition is only one environmental factor that can leave a long-lasting complex growth problem. Emotional deprivation also has a profound influence on the growth process and may interact with the provision of food [41]. A well-loved child is fed and nurtured, whereas a child with no prospect of a job or marriage or who is a burden may not be. Many of the subjects in this study were social outcasts, who dropped out of school. Females in particular were hidden away in their houses and only one female in the unoperated population married. Children need a good emotional climate to thrive. The mechanism of the effects of emotional deprivation on growth is not well documented but is linked to reduced growth hormone secretion and its associated growth failure.

10.4.2.2 Speech Implications

While facial growth in the unoperated subject presents without maxillary retrusion, unlike many operated patients, the speech outcomes for the same series of patients demonstrate almost unintelligible speech for the whole sample. This is illustrated in Fig. 10.16, which compares and contrasts the GOSLON results with the speech articulation outcomes for a group of UCLP subjects who had both dental study model and speech recordings.

Research on the Sri Lankan Cleft Lip and Palate archive has demonstrated that surgery, when delayed beyond 8 years of age and even earlier, results in permanent irremediable speech disorders [17, 49].

10.4.2.3 Racial Variation

The Sri Lankan Cleft Lip and Palate project was careful to use healthy control subjects from the Sri Lankan population. Sri Lankans present with bimaxillary protrusion, females on a skeletal 2 dental base and males on a mild skeletal 3 dental base. These are reflected within the cleft population. Interestingly, only one subject presented with a class II/2 dental arch relationship on a skeletal 2 dental base (Fig. 10.17).

In a recent (2003) project in Gujarat and Rajasthan in northern India, the vast majority of patients presenting had class II/2 dental arch relationships on skeletal 2 dental bases. This further emphasizes the need to have control subjects from the same racial grouping (Fig. 10.18).

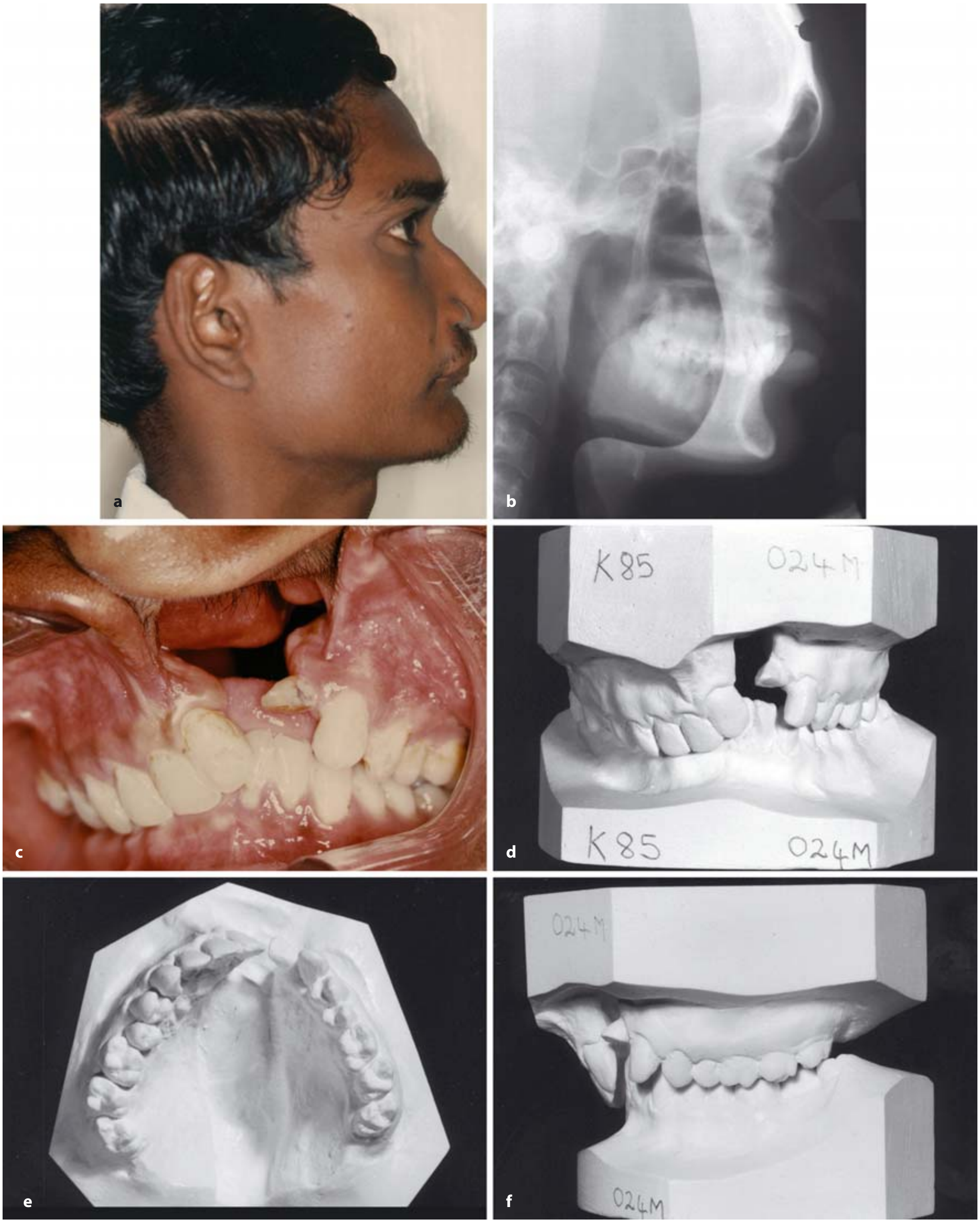

Fig. 10.17 a–f. Adult male subject with unoperated unilateral cleft of the lip and palate. This is a rare example of class II/2 arch relationship demonstrating retroclined upper incisors. Very broad upper arch and very low FM angle. The lower lip controls the upper labial segment causing retroclination

Fig. 10.18 a, b. This occlusion and facial pattern is characteristic of patients in northern India

10.4.2.4 Surgical Implications

It should not be inferred that because unoperated cleft lip and palate subjects grow relatively normally, surgical regimens delaying palatal surgery are indicated. This study does not examine the effects of surgery or the timing of that surgery upon facial growth outcomes.

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