European Journal of Orthodontics 19 (1997) 383–389

The craniofacial complex in karyotype 46,XY females

Kati Pietilä, Mathias Grön and Lassi Alvesalo

Department of Oral Development and Orthodontics, Institute of Dentistry, University of Oulu, Finland

SUMMARY The craniofacial cephalometric dimensions, angles and dimensional ratios of five Finnish individuals with complete testicular feminization (CTF) were compared with their first-degree relatives and population female and male controls. The linear and angular measurements were made from standardized lateral cephalograms of patients and normal population controls from the 'Kvantti Study' series.

The women with CTF tended to have cranial base and maxillary complex dimensions between those of the normal control females and males. Their mandibular corpus was found to be longer than in normal control females, while their ramus was shorter compared with that of normal males. They also showed a smaller sagittal length ratio of the maxilla to the mandible, a smaller ANB angle and a more acute gonial angle than in both normal control females and males. Comparison of the women CTF with their first-degree female relatives showed basically the same trends as when comparing them with normal female controls.

As the phenotype in these females with CTF is due to insensitivity to, or lack of androgens, it is suggested that the presence of the Y chromosome in these females leads to craniofacial dimensions between those of normal females and males which influences the growth of the mandibular corpus. This follows the same general metric pattern that is observed in many of their adult head and body dimensions as well as in their dental arches.

Introduction

Individuals with testicular feminization have the male karyotype 46,XY, although they are phenotypically females. The testicular feminization syndrome can be divided into two types: complete (CTF) and incomplete (ITF) forms (Dewhurst, 1971). The CTF disorder is estimated to occur in 1 in every 62 400 liveborn males (Jagiello and Atwell, 1962). Those with CTF have bilateral intra-abdominal or inguinal testes, blind-ended vagina and no Mullerian derivatives. The secondary sex characteristics are female, including normal breast development during puberty. The locus for the human cytosol androgen receptor is X-linked. The basic endocrine defect seems to be end-organ insensitivity to androgens, and the failure of androgen action plus testicular oestrogens could provide an explanation for the development of secondary female sex characteristics (Quigley *et al.,* 1992). Normal oestrogen action and even increased oestrogen production occur, which explains the hormonally induced female sex characteristics (MacDonald *et al.,* 1979). On the other hand, because of the androgen insensitivity, male characteristics in CTF individuals are presumed to be caused by the influence of the Y chromosome (Polani and Polani, 1979; Smith *et al.,* 1985).

The stature of 46,XY androgen-insensitive females was found to be above average for females and above that expected for their female midparental height. This supports the concept of a stature-determining role for Y-chromosomal genes, independent of their critical function in testicular determination (Quigley *et al.,* 1992; Smith *et al.,* 1985). Similar results have been reported after anthropometric studies on the same group of Finnish 46,XY women with CTF as in our report. Most of their body dimensions were found to be larger than those of female controls, but they were generally shorter than those of male controls. Their head circumference and length were larger than those of the female controls (Varrela *et al*., 1984). The size and shape of the craniofacial complex is found to be affected in individuals with sex-chromosome anomalies (Rzymski and Kosowicz, 1975; Peltomäki *et al*., 1989, Babić *et al*., 1991; Brown *et al*., 1993). Generally, an addition of the Y chromosome is found to increase linear dimensions.

In earlier studies of oral cavities in individuals with CTF, their permanent teeth were found to be larger in the labio-lingual dimension than the teeth of normal female controls as well as of their first-degree female relatives, and seemed to be closer in size to those of normal men (Alvesalo and Varrela, 1980). The enamel of the maxillary central permanent incisors had similar mesio-distal thickness in the CTF females as in the population control males and females, while the dentine was approximately the same thickness in the CTF females and control males and thicker than in control females (Alvesalo, 1985).

The aim of this study was to analyse the influence of the Y chromosome on size and shape of the craniofacial complex in 46,XY females with insensitivity to androgens.

Subjects and methods

Eight Finnish 46,XY females aged 18–42 years at the time of examination (mean age 25.4) with complete testicular feminization and four 46,XY females aged 9–29 (mean age 17.0) with more or less ambiguous genitalia were examined. The controls were eight first-degree female relatives aged 6–43 years (mean age 22.7), 45 normal females aged 9–56 years (mean age 24.6) and 46 normal males aged 9–43 years (mean age 26.3), taken from the same 'Kvantti Study' series of individuals with sex chromosomal abnormalities and normal control subjects as the 46,XY females. Most of the patients had had oestrogen therapy (not quantified), which had begun in early adulthood. Although oestrogen therapy is known to induce the pubertal growth spurt, it has been reported not to influence significantly final height (Kastrup, 1988).

The subjects were radiographed and a cephalometric analysis comprising linear and angular measurements was made from standardized lateral cephalograms. The reference points and planes used are shown in Figures 1 and 2. A sliding digital calliper (VIS, MAUa-E, Fabrik für feinmechanische Erzeugnisse 'General Swierczewski', 01–234, Warsaw, Poland) was used to measure distances between reference points (marked with a pencil on matte acetate film) to the nearest half millimeter. The angular measurements were made to the nearest whole degree with a protractor. Table 1 and Figures 1 and 2 show the variables used in the analysis. When there were two images of the structure, the reference point was placed in the midpoint between these images. The enlargement of the radiographs (8.7 per cent) was not corrected. The intra-observer error was analysed by a method suggested by Bland and Altman (1986). Lateral cephalograms of 20 patients attending the Dental Clinic at the University of Oulu were traced and measured twice.

The estimated error between the measurements was calculated using the formula:

$$\mathrm{SDd} = \sqrt{\Sigma (d_1 - d_2)^2 / 2n}$$

where ± 2 SDd are the limits within which 95 per cent of the differences between the repeated measurements are expected to lie; d_1 = first measurement; d_2 = second measurement; n = number of patients. The statistical comparisons, however, were made between those 46,XY females who had their first-degree relatives examined.

The error of measurement given in ±2 SD of the differences between the repeated measurements ranged from ±0.03 to ±0.36 (mean limit ±0.29) with the greatest error in the S–Cd dimension for linear measurments. Angular measurements varied between ±0.27 and ±0.67 (mean limit ±0.37) with the greatest error in the sph/man angle. Thus it was estimated not to be significant.

The craniofacial dimensions and plane angles of the five CTF women whose first-degree relatives were examined, were compared with their first-degree female relatives and with the mean of male and female population controls. In cases where a tooth was missing, the corresponding variable was omitted from the analysis of the dental and maxillary complex. All the subjects included in the analyses were over 16 years of age and had not lost their dentition.

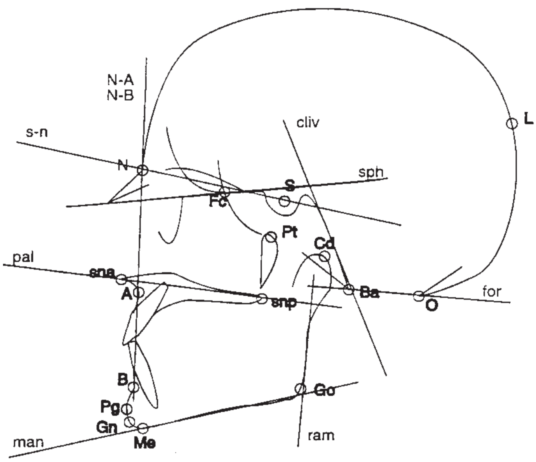

Figure 1 Reference points and planes used in the cephalometric analysis. Points: s = sella: the midpoint of sella turcica; N = nasion: the extreme anterior point on the frontonasal suture; sna = spina nasalis anterior: the extreme anterior point on the maxilla; snp = spina nasalis posterior: the extreme posterior point on the maxilla; Pt = pterygoid point: the extreme superior point of the pterygopalatine fossa; A = point A: the deepest point in the curvature of the maxillary alveolar process; B = point B: the deepest point in the curvature of the mandibular alveolar process; Pg = pogonion: the extreme anterior point of the chin; Me = menton: the extreme inferior point of the chin; Gn = gnathion: the midpoint between pogonion and menton; Go = gonion: the midpoint of the mandibular angle between ramus and corpus mandibulae; O = opisthion: the posterior border of foramen magnum; Ba = basion: the anterior border of foramen magnum; Cd = condylion: the extreme superior point of the condyle; Fc = fossa cranialis. the intersection between the sphenoidal plane and the larger wing of the sphenoid; L = lambda: the midpoint of the lambdoid suture on the external cranial contour. Planes: s–n = the sella–nasion line; sph = the sphenoidal plane; cliv = the clival plane; for = the foramen magnum plane; pal = the palatal plane (sna–snp); occ = the occlusal plane (see Figure 2: from the intersection of the upper and lower incisors to the occlusal contact of the upper and lower first molars); man = the mandibular plane (a tangent to the lower border of the mandible); ram = the ramal plane (a tangent to the dorsal surface of the ramus with exclusion of the condyle); N–A = the nasion–A line; N–B = the nasion–B line.

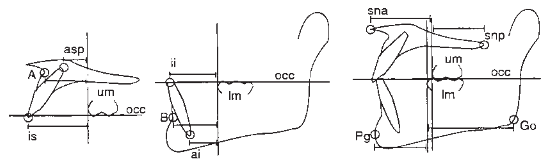

Figure 2 Occlusal analysis reference points from which measurements were made parallel to the occlusal plane: is = incision superius: the tip of the crown of the most anterior maxillary central incisor; asp = apex superius: the root apex of the most anterior maxillary central incisor; um = upper first molar: its most mesial point; ii = incision inferius: the tip of the crown of the most anterior mandibular central incisor; ai = apex inferius: the root apex of the most anterior mandibular central incisor; lm = lower first molar: its most mesial point; occ = the occlusal plane (from the intersection of the upper and lower incisors to the occlusal contact of the upper and lower first molars); A = point A: the deepest point in the curvature of the maxillary alveolar process; B = point B: the deepest point in the curvature of the mandibular alveolar process; Pg = pogonion: the extreme anterior point of the chin; Go = gonion: the midpoint of the mandibular angle between ramus and corpus mandibulae

Results

Data for every CTF individual having smaller linear dimensions and dimensional ratios than the mean male and female population controls, smaller values than their first-degree female relatives, and larger mean values than those of the control females are shown in Table 1.

In the cranial base dimensions and in the distances from the cranial base to the maxillary complex all but one of the CTF women's means fell between those of the normal control females and males. Some values were even greater than those of the control males. In the maxillary complex most of the CTF women's mean values (4/6) fell between those of the normal control females and males, the Go–Pg being longer in the CTF women than in normal control females. The CTF women also had a shorter ramus compared with that of normal males.

In the dental complex the CTF women presented larger mean sagittal dimensions than both normal females (9/10) and males (7/10). When comparing the sagittal length ratio of the maxilla to the mandible, the CTF women showed a smaller value than those of the normal females and males. This was supported by CTF females tending to have a shorter maxilla and a longer mandible in proportion to the anterior cranial base than both normal females and males. The ANB and the gonial angle of the CTF women also appeared smaller than in both normal control females and males.

The values for individual CTF females showed a strong tendency to exceed the mean of control males and females for the cranial base dimensions, the length of the mandibular corpus and

Table 1 Cephalometric dimensions (in mm), dimensional ratios and angles of the women with complete testicular feminization (CTF), their first-degree female relatives and controls of both sexes.

	CTF		Female relatives		Control females		Control males		CTF < $\overline{X3}$ (ratio)	CTF < female rel. (ratio)	CTF $\overline{X}$ > control females $\overline{X}$ (ratio)
	Mean	(*n*/SD)	Mean	(*n*/SD)	Mean	(*n*/SD)	Mean	(*n*/SD)			
Age	24.85	5/9.65	24.8	5/10.58	25.35	35/7.41	27.39	30/6.8			
LINEAR DIMENSIONS											
Cranial base											
S–N	75.6	5/1.43	70.4	5/2.9	73.11	35/3.17	76.55	30/3.29	2/5	1/5	
S–Fc	28.70	5/3.87	27.5	5/2.52	27.2	35/2.54	28.9	30/2.65	2/5	1/5	
Fc–N	46.6	5/3.07	42.7	5/4.83	45.74	35/3.53	47.35	30/3.76	2/5	1/5	
S–Ba	47.1	5/2.84	47.3	5/3.6	45.37	35/2.66	48.47	30/3.08	2/5	3/5	
Ba–Pt	53.9	5/0.42	52.9	5/3.6	52.04	35/3.39	53.08	30/4.17	0/5	2/5	
Fc–Pt	20.1	5/3.93	18.1	5/3.49	17.73	35/2.66	19.95	30/3.03	2/5	2/5	
S–L	118.2	5/4.51	116.3	5/3.05	114.89	35/5.56	118.65	30/5.35	3/5	1/5	
O–Ba	36.1	5/2.88	34.0	5/2.35	32.94	35/3.06	36.15	30/3.31	1/5	1/5	8/8
Cranial base to maxillary complex											
S–Cd	25.3	5/3.67	21.8	5/2.64	23.06	35/3.03	25.27	30/2.87	2/5	0/5	
O–Cd	52.9	5/2.38	53.1	5/3.29	51.0	35/3.8	54.1	30/3.85	2/5	3/5	
O–Go	62.1	5/6.67	57.0	5/1.32	62.31	35/5.25	68.27	30/7.83	3/5	1/5	
S–Go	79.7	5/4.21	73.3	5/4.38	76.76	35/5.62	87.22	30/6.42	3/5	0/5	
N–Me	127.2	5/5.64	121.2	5/5.69	119.91	35/6.1	130.6	30/7.16	1/5	0/5	
N–sna	55.0	5/1.97	53.8	5/1.3	53.31	35/2.82	57.7	30/3.18	3/5	1/5	
S–sna	90.2	5/2.25	86.9	5/2.95	89.14	35/4.43	94.25	30/4.32	4/5	1/5	
S–snp	52.8	5/0.91	48.4	5/0.82	49.91	35/2.72	53.35	30/3.56	0/5	0/5	7/8
Maxillary complex											
sna–Me	73.1	5/3.85	69.5	5/6.51	68.31	35/5.24	74.33	30/5.76	1/5	1/5	
sna–snp	53.3	5/2.44	52.0	5/2.89	53.96	35/2.87	56.65	30/3.39	4/5	2/5	
A–snp	48.5	5/2.72	47.1	5/2.97	49.19	35/2.7	51.93	30/3.32	3/5	3/5	
Go–Pg	87.5	5/4.34	79.5	5/7.13	78.41	35/5.28	82.45	30/4.98	0/5	0/5	
Cd–Go	57.9	5/5.16	54.4	5/3.52	56.79	35/4.16	65.22	30/5.47	4/5	0/5	
Cd–Gn	124.7	5/6.44	116.9	5/6.48	117.4	35/6.5	127.55	30/6.63	2/5	1/5	4/6
Dental complex											
is–um	29.88	4/2.56	29.1	5/3.54	28.06	33/3.44	28.64	29/2.73	1/4	2/4	
asp–um	19.13	4/2.75	18.1	5/3.03	17.21	33/3.4	18.29	29/3.17	1/4	2/4	
A–um	21.25	4/3.71	20.8	5/4.72	20.79	33/3.06	21.97	29/3.11	2/4	2/4	
ii–lm	25.13	4/2.75	24.6	5/1.82	24.72	32/2.61	23.81	29/3.68	2/4	2/4	
ai–lm	16.13	4/5.63	13.3	5/2.77	14.81	32/4.01	15.14	29/4.17	2/4	2/4	
B–lm	20.88	4/4.96	18.2	5/3.46	20.45	32/3.84	20.39	28/4.04	2/4	1/4	
sna–um	25.75	4/5.14	25.3	5/4.19	25.36	33/3.44	26.74	29/3.74	3/4	2/4	
snp–um	26.63	4/5.44	26.1	5/2.79	28.12	33/2.95	30.45	29/5.57	3/4	1/4	
Pg–lm	25.13	4/7.33	21.5	5/3.48	23.47	32/4.8	24.5	29/5.09	3/4	1/4	
Go–lm	60.38	4/5.12	55.4	5/6.28	52.89	32/6.09	55.76	29/5.45	0/4	0/4	9/10

of the maxillary dental complex, and also for the the sagittal length ratio of the mandibular corpus to the anterior cranial base. The trend was reversed in the length of the mandibular ramus, the ANB and the gonial angle, and in the sagittal length ratio of the maxilla to the mandible and to the anterior cranial base.

Comparison of the women with CTF with

Table 1 *continued*

	CTF		Female relatives		Control females		Control males		CTF < $\overline{X3}$ (ratio)	CTF < female rel. (ratio)	CTF $\overline{X}$ > control females $\overline{X}$ (ratio)
	Mean	(*n*/SD)	Mean	(*n*/SD)	Mean	(*n*/SD)	Mean	(*n*/SD)			
DIMENSIONAL RATIOS											
sna–snp/ Go–Pg	0.61	5/0.03	0.66	5/0.06	0.69	35/0.05	0.69	30/0.05	5/5	4/5	
sna–snp/ S–N	0.71	5/0.03	0.74	5/0.02	0.74	35/0.04	0.74	30/0.04	4/5	3/5	
Go–Pg/ S–N	1.16	5/0.06	1.13	5/0.08	1.07	35/0.07	1.08	30/0.07	0/5	3/5	
S–Go/ N–Me	0.63	5/0.03	0.61	5/0.05	0.64	35/0.05	0.67	30/0.04	3/5	1/5	
N–sna/ sna–Me	0.75	5/0.04	0.78	5/0.08	0.78	35/0.07	0.78	30/0.07	4/5	3/5	1/5
ANGLES											
Cranial base											
sph/S–N	14.0	5/6.16	14.6	5/3.21	16.94	35/5.87	13.53	30/5.16	3/5	2/5	
sph/cliv	113.8	5/8.23	111.2	5/3.03	111.0	35/6.9	110.27	30/6.6	2/5	2/5	
for/cliv	126.0	5/9.54	123.6	5/3.05	127.94	35/5.27	124.0	30/6.07	3/5	3/5	1/3
Maxillary complex sagittal											
SNA	80.6	5/2.07	81.6	5/2.07	82.83	35/4.42	82.67	30/4.08	4/5	3/5	
SNB	78.8	5/2.17	77.4	5/2.97	78.91	35/3.44	79.8	30/3.83	4/5	2/5	
ANB	1.8	5/2.05	4.2	5/2.28	3.91	35/3.49	2.87	30/2.7	4/5	4/5	0/3
Maxillary complex vertical											
pal/occ	9.0	4/3.46	7.8	5/4.27	7.85	34/3.72	5.73	30/3.6	1/4	1/4	
pal/man	25.8	5/4.66	25.6	5/4.83	23.94	35/5.26	21.9	30/6.33	1/5	2/5	
man/occ	15.75	4/1.71	18.0	5/2.35	15.88	34/4.46	16.0	30/4.46	2/4	3/4	
man/ram	111.8	5/5.12	116.4	5/4.04	117.54	35/8.83	116.77	30/7.17	4/5	4/5	
man/s–n	31.8	5/2.86	34.8	5/4.76	30.89	35/6.04	29.57	30/6.7	2 /5	3/5	
sph/man	18.0	5/8.34	20.0	5/4.42	13.77	35/6.0	15.8	30/8.57	1/5	1/5	
for/man	30.2	5/6.8	32.2	5/4.6	30.31	35/6.1	29.63	30/8.8	3/5	2/5	4/7

$\overline{X3}$: mean of control females and control males.
CTF < $\overline{X3}$ (ratio): how many times the value of the CTF < $\overline{X3}$/comparison.
CTF < female relatives (ratio): how many times the value of the CTF < subjects' female relative/comparison.
CTF $\overline{X}$ > control females $\overline{X}$ (ratio): in how many variables the mean of the CTFs > mean of control females/variables.

their first-degree female relatives showed basically the same trends concerning the cranial base, the distances from the cranial base to the maxillary complex, the maxillary complex, the dental complex and the dimensional ratios, as when comparing them with normal female controls (see Table 1).

Discussion

This is to our knowledge the first report on the dimensions and form of the craniofacial complex in 46,XY females with CTF. The results from tooth crown studies have shown that the Y chromosome affects both dentine and enamel growth, which is probably due to the proliferative activity of odontoblasts and to the secretory activity of ameloblasts (Alvesalo and Tammisalo 1981; Alvesalo, 1985; Alvesalo *et al.*, 1987, 1991). Assuming pleiotropy (i.e. a gene having more than one phenotypic effect), the larger craniofacial dimensions in the CTF women may thus be the consequence of the Y chromosome genes affecting proliferative and appositional growth in the craniofacial complex, independently of androgen action. These effects on the craniofacial dimensions may be caused by increased proliferative growth in the condyle, in the sutures and synchondroses of the cranial base and the maxilla and by increased intramembranous apposition. This increased growth might be followed by an alteration of the mandibular morphology, or alternatively by compensatory changes in the mandibular growth pattern.

Generally, the results indicate that the shape of the cranial base in CTF females is not markedly affected and resembles that of normal controls. The dimensions of the cranial base, and of the maxillary and dental complex in the CTF patients fall between those of normal females and males. This finding of larger size is in agreement with those of Varrela *et al*. (1984) of larger body dimensions, head circumference, and length of head and face in women with CTF compared with normal women. The findings are also in agreement with Alvesalo and Varrela (1980) that the permanent teeth of 46,XY females are similar in size to those of normal men, and with the findings of Grön and Alvesalo (1997) of larger dental arch dimensions in CTF subjects compared with normal women. In this study CTF females presented a longer mandibular corpus than normal females, with values even exceeding those of normal males. They also had a shorter ramus than normal males, resembling normal females in this dimension. Their gonial angles are more acute than in all control subjects, which might be a sign of compensation for the overgrowth of the corpus to preserve the harmony of the lower face. The sagittal length ratio of the maxilla to the mandible was smaller in those CTF subjects than in normal females and males, and this trend was confirmed by the maxilla being shorter and the mandible longer in proportion to the anterior cranial base. As would be expected, those CTF females also presented an ANB angle smaller than both normal control females and males. Due to the small sample size and missing values, no statistically significant differences could be shown with a non-parametric test, which otherwise would have been appropriate. These results are in concordance with the findings by Grön and Alvesalo (1997) of females with CTF having a more mesial molar and canine occlusion compared with normal females, and an increased vertical overbite, which might be explained by the acute gonial angle.

Conclusion

As the phenotype in females with CTF is due to insensitivity to, or lack of proper androgens, it is suggested that the presence of the Y chromosome in the females in this study leads to craniofacial dimensions falling between those of normal females and males and especially influencing the growth of the mandibular corpus. This follows the same general metric pattern, which is observed in many of their adult head and body dimensions as well as in their dental arches.

Address for correspondence

Dr Mathias Grön
Department of Oral Development and Orthodontics
Institute of Dentistry
University of Oulu
Aapistie 3
90220 Oulu
Finland

Acknowledgements

We wish to thank associate Professor Juha Tienari and graduate student Jouko Remes from the Department of Applied Mathematics and Statistics at the University of Oulu for statistical treatment of the data. The study was supported by the Academy of Finland and the University of Turku Foundation.

References

Alvesalo L 1985 Dental growth in 47,XYY males and in conditions with other sex-chromosome anomalies. In: Sandberg A A (ed.) The Y chromosome, Part B: Clinical aspects of Y chromosome abnormalities. Alan R Liss, New York, pp. 277–300

Alvesalo L, Tammisalo E 1981 Enamel thickness in 45,X females' permanent teeth. American Journal of Human Genetics 33: 464–469

Alvesalo L, Varrela J 1980 Permanent tooth sizes in 46,XY females. American Journal of Human Genetics 32: 736–742

Alvesalo L, Tammisalo E, Therman E 1987 47,XXX females, sex chromosomes, and tooth crown structure. Human Genetics 77: 345–348

Alvesalo L, Tammisalo E, Townsend G 1991 Upper central incisor and canine tooth crown size in 47,XXY males. Journal of Dental Research 70: 1057–1060

Babić M, Mićić M, Jaksić N, Mićić S 1991 An extra X chromosome effect on craniofacial morphogenesis in men. European Journal of Orthodontics 13: 329–332

Bland J M, Altman D G 1986 Statistical methods for assessing agreement between two methods of clinical measurement. Lancet i: 307–310

Brown T, Alvesalo L, Townsend G C 1993 Craniofacial patterning in Klinefelter (47 XXY) adults. European Journal of Orthodontics 15: 185–194

Dewhurst C J 1971 The XY female. American Journal of Obstetrics and Gynecology 109: 675–688

Grön M, Alvesalo L 1997 Dental occlusion and arch size and shape in karyotype 46,XY females. European Journal of Orthodontics 19: 329–335

Jagiello G, Atwell J 1962 Prevalence of testicular feminization. Lancet i: 329–329

Kastrup K W 1988 Oestrogen therapy in Turner's syndrome. Acta Paediatrica Scandinavica (Suppl) 343: 43–46

MacDonald P C, Madden J D, Brenner P F, Wilson J D, Siiteri P K 1979 Origin of estrogen in normal men and in women with testicular feminization. Journal of Clinical Endocrinology and Metabolism 49: 905–916

Peltomäki T, Alvesalo L, Isotupa K 1989 Shape of the craniofacial complex in 45,X females: cephalometric study. Journal of Craniofacial Genetics and Developmental Biology 9: 331–338

Polani P E, Polani N 1979 Dermatoglyphics in testicular feminization syndrome. Annals of Human Biology 6: 417–430

Quigley C A *et al.* 1992 Complete deletion of the androgen receptor gene: definition of the null phenotype of the androgen insensitivity syndrome and determination of carrier status. Journal of Clinical Endocrinology and Metabolism 74: 927–932

Rzymski K, Kosowicz J 1975 The skull in gonadal dysgenesis—a roentgenometric study. Clinical Radiology 26: 379–384

Smith D W, Marokus R, Graham J M 1985 Tentative evidence of Y-linked statural gene(s)—growth in the testicular feminzation syndrome. Clinical Pediatrics 24: 189–192

Varrela J, Alvesalo L, Vinkka H 1984 Body size and shape in 46,XY females with complete testicular feminization. Annals of Human Biology 11: 291–301