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Mandibular growth pattern in Turner's syndrome

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SUMMARY In a group of 15 women with 45,X chromosome constitution, mandibular growth type was investigated by using both linear and angular measurements. The sum of the saddle, articular and gonial angle, lower gonial angle and *y*-axis was significantly greater. In addition the posterio-anterior facial height ratio in women with Turner's syndrome was significantly smaller than in the controls (61 women with 46,XX chromosome constitution), indicating a tendency to backward and downward growth changes in the mandible, caused by an X chromosome deficiency.

Introduction

Turner's syndrome is a relatively common disorder that occurs in 1:2500 female births and is caused by complete or partial absence of one of the X chromosomes (Evans, 1977; Hook and Warburton, 1982). Although a large majority of patients with Turner's syndrome are characterized by loss of a complete X chromosome, there are also rare cases with structurally abnormal X chromosomes, such as X_p or X_q deletion, isochromosome X_q ring X chromosome, which all exhibit a number of characteristic features of this syndrome (Wyss *et al*., 1982).

Growth-related anomalies represent part of the most important effects of X chromosome deficiency. In addition to short stature, which is one of the main characteristics of Turner's syndrome, cranial growth reduction and decreased mesiodistal dimensions of permanent teeth have also been registered (Shimaguchi *et al*., 1961; Filipsson *et al*., 1965; Park, 1977). Likewise, a prolonged growth period caused by slowing down of the epiphyseal cartilage fusion and early eruption of permanent teeth indicates changes of growth timing which are the result of an X chromosome deficiency (Acheson and Zampa, 1961; Filipsson *et al*., 1965).

Since it has been shown (Jensen, 1985) that women with Turner's syndrome exhibit a flattened cranial base, bimaxillary retrognathism and a posteriorly inclined mandible, this study was designed to investigate the role of this aberration in the control of mandibular growth rotation.

Subjects and methods

Fifteen female patients with Turner's syndrome, 45,X chromosome constitution, aged from 24 to 37 years, entered the study. From the lateral cephalogram tracings, using Björk's method (1969), adapted and modified by Jarabak and Fizzel (1972), mandibular growth rotation type has been determined. The parameters used for the analysis are presented in Figure 1. The accuracy of the linear and angular measurements was 0.5 mm and 0.5 degrees respectively, without correction for linear enlargement. Sixty-one female volunteers, dental students aged 23–28, served as controls. Dahlberg's method was used to test the reliability of the measurements, and Student's *t*-test was used to measure the differences between the observed groups.

Results

Comparative cephalometric analysis between women with 45,X karyotype and women with 46,XX karyotype revealed a tendency to backward and downward growth changes in the mandible in women with an X chromosome deficiency. This could be supported by all of the relevant parameters examined (Tables 1 and 2). With regard to Björk's criteria, an anterior and

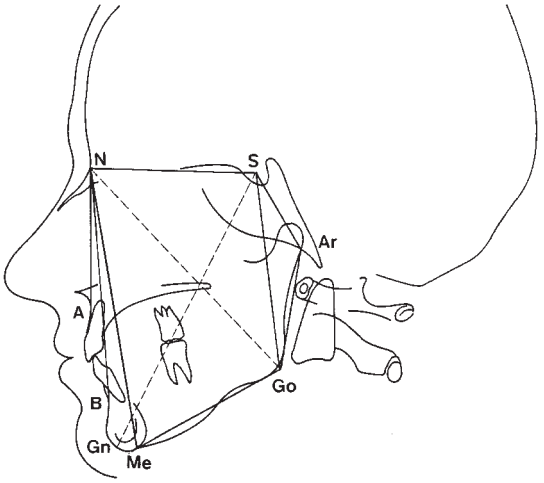

Figure 1 Parameters used for the analysis: NMe, anterior facial height; SGo, posterior facial height; NSAr, saddle angle; SArGo, articular angle; ArGoMe, gonial angle; NGoMe, lower gonial angle; NSGn, *y*-axis; SNA, maxillary prognathism; SNB, mandibular prognathism; ANB, sagittal jaw relationship.

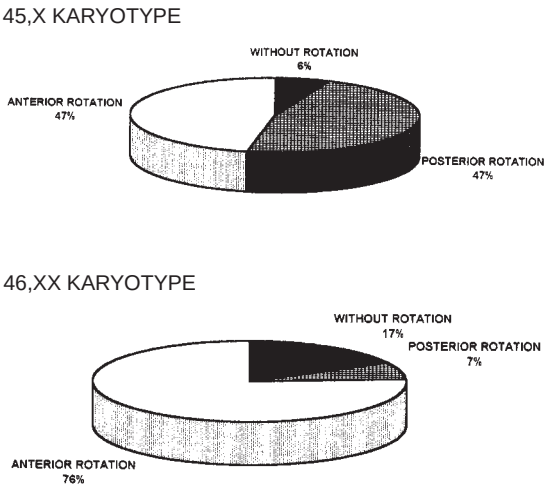

Figure 2 Distribution of mandibular growth rotation in women with 45,X and 46,XX karyotype.

posterior rotation was equally expressed in the Turner's syndrome group, while an anterior rotation significantly prevailed in the controls (Figure 2).

Facial polygon

The sum of the saddle, articular and gonial angles in women with Turner's syndrome was found to be larger than those of the controls (Table 2 and Figure 3) and the difference was statistically significant ($P < 0.001$). However, the articular angle measurements were not statistically significantly different between the observed groups.

Both the lower gonial angle and *y*-axis were larger by approximately 3–4 degrees in women with 45,X karyotype compared with those of the controls.

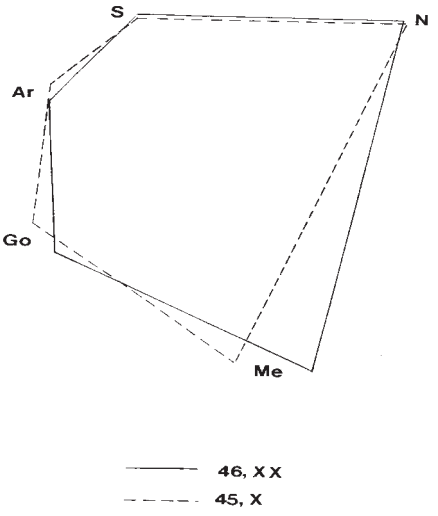

Figure 3 Superimposition of facial polygon in women with 45,X and 46,XX karyotype.

Posterio-anterior facial height ratio

Unlike anterior facial height, the posterior facial height was significantly reduced in women with Turner's syndrome, by approximately 9 per cent. Thus, the posterio-anterior facial height ratio significantly differed between the groups by about 4 per cent.

Sagittal jaw relationship

Both maxilla and mandible were retrognathic. SNA and SNB angles were significantly decreased nearly to the same extent, approximately 5 degrees, in the syndrome group. There was no significant difference in sagittal jaw relationship between the groups.

Table 1 Linear measurements

Parameters	45,X	46,XX	*t*	Error of measurement
Anterior facial height (mm)	119.2 ± 7.3	122.4 ± 6.1	NS	0.80
Posterior facial height (mm)	77.6 ± 6.0	84.6 ± 5.5	***	0.72
Postero-anterior facial height ratio (%)	65.2 ± 5.2	69.1 ± 4.4	**	0.15

Level of significance according to *t*-test: ***$P < 0.001$; **$P < 0.01$; *$P < 0.05$.

Table 2 Angular measurements

Parameters	45,X	46,XX	*t*	Error of measurement
Saddle angle (NSAr)	129.9 ± 8.5	126.3 ± 5.5	*	0.75
Articular angle (SArGo)	142.2 ± 13.8	142.9 ± 6.4	NS	0.66
Gonial angle (ArGoMe)	125.2 ± 8.1	121.7 ± 5.5	*	0.81
Facial polygon	397.2 ± 7.5	390.7 ± 6.3	***	0.20
Lower gonial angle (NGoMe)	74.7 ± 5.7	71.9 ± 3.8	*	0.50
y-axis (NSGn)	71.9 ± 5.9	67.9 ± 4.0	**	0.78
SNA	75.2 ± 2.5	81.4 ± 3.6	***	0.37
SNB	73.1 ± 4.6	78.7 ± 3.6	***	0.73
ANB	2.1 ± 3.2	2.7 ± 2.4	NS	0.10

Level of significance according to *t*-test: ***$P < 0.001$; **$P < 0.01$; *$P < 0.05$.

Discussion

Although Björk's method is considered insufficiently reliable in facial growth prediction, in adults where growth processes have already finished, it may serve in evaluation of the prevailing type of growth changes in the mandible (Björk, 1969). However, this method is unable to detect possible directional changes in mandibular growth rotation during craniofacial complex development.

On the basis of the results obtained it was ascertained that an X chromosome deficiency produced an increasing tendency to backward growth changes in the mandible. Although, there was no evidence concerning mandibular growth changes in patients with different chromosomal anomalies, some finding such as a posterior inclined mandible in Turner's syndrome, reported by Jensen (1985), lead to the same conclusion.

Likewise, this chromosomal anomaly reduced cranial growth capacity, but growth inhibition rates of the craniofacial structures examined were found to be different. Unlike anterior facial height, posterior facial height was significantly decreased in women with 45,X karyotype, affecting alteration in the usual posterio-anterior facial height ratio. It is uncertain whether reduced growth capacity of facial height caused by an X chromosome deficiency has any influence on the direction of mandibular growth rotation, or whether an underdeveloped posterior facial height represents just a consequence of backward growth changes in the mandible.

Bimaxillary retrognathism, as well as a skeletal Class I jaw relationship was also registered. These features could be accompanied by a decreased saddle angle, but also with a similar degree reduction of both the maxilla and mandible (Babić *et al*., 1993).

Evidence concerning two patients with Turner's syndrome, having both decreased anterior and posterior facial height, but greater posterio-anterior facial height ratio than in the controls, suggests that cranial growth inhibition

does not always coincide with posterior rotation. Two patients with atypical facial morphology for Turner's syndrome in this sample can be explained by the fact that facial morphology, like most morphological characteristics, probably has polygenic control (Sussane, 1975). Variation of these characteristics is closely related with an increased susceptibility toward genetic changes and environmental factors.

With regard to our findings, X chromosome genes seem to have an important influence on mandibular growth rotation. Prevalence of forward growth changes in the mandible in women with normal chromosomal constitution and equally distributed forward and backward growth changes in the group of women with an X chromosome deficiency, indicates that loss of a single X chromosome gene affects the direction of mandibular growth rotation, by stimulating clockwise growth changes.

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