

# Root length, crown height, and root morphology in Turner syndrome

Marit Midtbø and Agnar Halse

Department of Orthodontics and Facial Orthopedics and Department of Oral Radiology, School of Dentistry, University of Bergen, Bergen, Norway

Midtbø M, Halse A. Root length, crown height, and root morphology in Turner syndrome. *Acta Odontol Scand* 1994;52:303–314. Oslo. ISSN 0001-6357.

Root length, crown height, and root morphology were studied on intraoral and panoramic radiographs in 33 Turner syndrome patients aged 7.0–16.7 years, subdivided on the basis of karyotype. Thirty-three normal girls aged 10.2–16.4 years served as controls. In the 45X patients and, with the exception of a few teeth, also in the isochromosome and mosaic karyotypes, root length and crown height of incisors, canines, and premolars were significantly reduced. Some teeth showed altered crown–root proportions. Maxillary first premolars showed a significantly increased number of two-rooted and three-rooted variants. Mandibular premolars and molars had a complex root morphology, and a classification system was established including four premolar and six molar root types. Premolars had a significantly increased number of root components. Some of the variants, such as a molar-like second premolar, are apparently specific for these patients. On several first molars a radix entomolaris was identified. Two separate mesial and one or two separate distal roots were also frequently seen. Our investigation demonstrates that X-chromosome deficiency influences root formation.

□ *Anatomy; tooth; X chromosome*

Marit Midtbø, School of Dentistry, Årstadveien 17, N-5009 Bergen, Norway

Turner syndrome is a sex chromosomal disorder associated with a female phenotype. A chromosome examination of 34,910 liveborn Danish children showed a prevalence of 1 per 2130 girls (1). The main symptoms are growth deficiency and sexual infantilism, but various somatic abnormalities have been reported as a part of the syndrome. The girls are differently affected; the only ubiquitous characteristic is shortness of stature (2, 3).

Several karyotypes responsible for the syndrome have been identified, the most common being monosomy X, found in about 50% of the girls. Mosaics (45X/46XX) and isochromosome for the long arm of X (46X,i(Xq)) are found in about 24% and 17% of the cases, respectively (4). Rarer conditions such as ring X have also been observed.

The degree of X-chromosome deficiency is considered to influence the phenotype. The least affected clinical forms are almost invariably associated with a partial loss of the X chromosome. The opposite is not necessarily true; partial sex chromosome

monosomies may be associated with the typical clinical picture found in 45X patients (5).

The craniofacial morphology shows some specific alterations. The cranial base is flattened, the jaws are more posteriorly positioned, and a retrognathic face type is common. The mandibular alveolar arch has been reported as broad and short and the maxillary arch as narrow (6–8). The prevalence of lateral crossbites is high (9, 10).

Tooth crown size and morphology are also influenced; the tooth crown is smaller and the morphology simplified (11–17). Some reports conclude that the changes in tooth crown size and morphology differ for the various karyotypes (14), whereas others have found no significant differences (17).

The roots have been reported to be shorter than normal (18). In a material of 45X patients a significantly higher number of mandibular premolars with separate root canals has been found (19), which indicates that root morphology is influenced by X-chromosome monosomy. A more detailed knowledge of the root morphology in the

Table 1. Patients distributed on the basis of age and karyotype

| Karyotype      | n  | Age (years) |      |
|----------------|----|-------------|------|
|                |    | Range       | Mean |
| Monosomy X     |    |             |      |
| 45X            | 24 | 7.0–16.7    |      |
| Mosaics        |    |             |      |
| 45X/46XX       | 3  | 12.5–15.3   |      |
| 45X/46XY       | 1  | 14.7        |      |
| 45X/46X,i(Xq)  | 1  | 12.8        |      |
| 45X/46X,r(Xq)  | 1  | 15.8        |      |
| Isochromosomes |    |             |      |
| 46X,i(Xq)      | 3  | 8.7–12.8    |      |
| Total          | 33 | 7.0–16.7    | 12.1 |
| Controls       | 33 | 10.2–16.4   | 12.7 |

different karyotypes of the syndrome is not available.

The aim of the present investigation was to analyze 1) root length, 2) tooth proportions expressed as the ratio between root length and crown height, and 3) root morphology. Possible differences between the karyotypes were also investigated.

## Materials and methods

This investigation is part of a systematic study of Turner syndrome patients whose intention is to evaluate growth and development before, during, and after therapy with growth hormone and estrogen. The karyotyping was done by chromosome analysis of peripheral lymphocytes. The karyotyping, the hormone therapy, and the study of general variables were performed by the Department of Pediatrics, University of Bergen.

The material comprised 33 patients aged 7.0–16.7 years, from different parts of Norway. The distribution of the patients on the basis of karyotype and age is shown in Table 1. Before start of hormone treatment the patients were examined clinically at the Department of Orthodontics and Facial Orthopedics. At the Department of Oral Radiology a series of intraoral radiographs

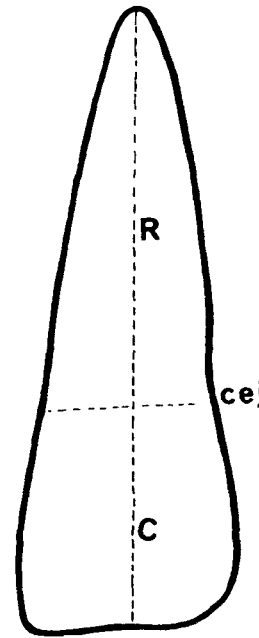


Fig. 1. The method of measuring on radiographs illustrated schematically. The line connecting the mesial and distal cemento-enamel junction, cej, as seen on the outer contour, intersects the long axis of the tooth, thereby defining the root length (R) and the crown height (C).

were taken with a paralleling technique. As evaluated by Larheim & Eggen (20), this technique gives about 5% magnification. A panoramic radiograph was taken with an orthopantomograph, model OP5 (Palomex Instrumentarium Corp., Helsinki, Finland), which, according to Thanyakarn et al. (21), gives a vertical magnification of 13%–20% of the premolars.

As controls served 33 girls aged 10.2–16.4 years from the files of the Department of Orthodontics. None of them had a known history of genetic or hormonal disorders. For each of these patients a panoramic radiograph and a series of intraoral radiographs were available.

## Measurements of incisors and canines

Root length and crown height were measured on intraoral radiographs. Measurements, as illustrated in Fig. 1, were per-

formed by use of a magnifying lens (7×) with 0.1-mm gradation and a coordinate system. To describe the proportions of the tooth type, the crown-root index (CRI) was calculated as root length divided by crown height (22). Only teeth with complete root formation were measured. Teeth with markedly curved roots, severe rotations, or extensive restorations were excluded. Five patients under treatment with fixed orthodontic appliances were not measured. Of 394 incisors and canines, 234 were measured.

The measurements were repeated after 1 month, and the measurement error calculated. The error for a single measurement averaged 0.16 mm, which was considered to be acceptable. The mean of the two measurements was used in the further calculations. No correction was made for enlargement.

#### *Measurements of premolars*

The premolars were measured on panoramic radiographs. The same procedures as on the intraoral pictures were used for measuring, exclusion, and calculation. Of 260 premolars, 139 were measured. The error of a single measurement was on average 0.21 mm, which was considered acceptable.

#### *Root morphology*

Root morphology was studied by means of intraoral and panoramic radiographs, and every tooth could be viewed in at least two different projections. Initially, all roots were identified, but only roots with either closed or nearly closed apices were included. Of 917 teeth, 656 were scored. Third molars were excluded.

A pilot study of the material showed large variation in the root morphology of premolars and mandibular molars, and a classification system had to be established. The nomenclature was based on Carlsen's *Textbook of Dental Morphology* (22). The following variables were used in the identification: a) the number of root canals, b) the number of apices, c) the outline of the root surface, and d) the location of the bifurcation and the degree of separation.

The maxillary premolars were classified into three groups: a) premolar with non-separate root and apex; b) premolar with two separate root components located buccally and lingually; degree of separation >0.3; and c) premolar with three separate root components: two buccally and one lingually positioned; degree of separation >0.3.

Four mandibular premolar root types were defined. Type 1: one-rooted premolar with one root canal and one apex; type 2: one-rooted premolar with separate root canals and one apex; type 3: premolar with two or more separate buccal and lingual root components; and type 4: two-rooted premolar with separate mesial and distal root components.

Six mandibular molar roots types were defined: type 1: one-rooted molar with non-separate mesial and distal root components; type 2: two-rooted molar with separate mesial and distal root components; type 3: three-rooted molar with separate mesial, distal, and distolingual root components; the supernumerary distolingual root component identified as radix entomolaris; type 4: three-rooted variant with separate mesial and two separate distal root components; type 5: three-rooted variant with two separate mesial and one separate distal root component; and type 6: variant with separate mesial and distal root components of which either the mesial or the distal, or both, have greater width than normal and separate apices.

The classification of all teeth on the basis of root morphology was done by the two observers jointly, one of them a specialist in oral radiology (A. Halse). The investigators were not aware of the patient's karyotype during the registrations.

#### *Taurodontism*

Molars and premolars were investigated for taurodontism as proposed by Shaw (23) and Madeira et al. (24). Both panoramic and intraoral radiographs were used. The same premolars and molars as in the root morphology part were included and scored by the two observers.

### Statistics

Measurement error was calculated from the formula  $\tau^2 = \frac{d^2}{2n}$  (25), where  $d$  is the difference between the first and second measurement for each variable. Differences in crown height, root length, and CRI between the teeth in the left and right side were tested by paired  $t$  test. As no significant differences were found, only the right side was used and the further calculations.

The results of the tooth measurements were subdivided into three groups on the basis of the patients' karyotype. The sample sizes were small, so Wilcoxon's sign rank test was used and the following comparisons done for all the variables: a) 45X patients versus controls; b) isochromosome and mosaics in one group versus controls; and c) isochromosome and mosaics in one group versus 45X patients.

For differences in morphology the chi-square test was used. The comparisons were made between Turner patients and controls. Regrouping was done before the following testing: a) The number of one-rooted maxillary premolars was compared with the number with two and three separate root components; b) The number of mandibular premolars with non-separated roots and one root canal (type 1) was compared with the number with deviating morphology (types 2–4); and c) the number of mandibular molars with one or two roots (types 1 and 2) was compared with those with deviating anatomy (types 3–6).

All calculations were performed by means of a computer program (26).

## Results

### 45X patients

Both in the mandible and in the maxilla the crown height of incisors and canines was significantly reduced compared with that in normal controls (Table 2). Root length was also significantly reduced, except for the mandibular canine. Percentage size reduction ranged from 4.5 to 14.0, the largest reduction found for root length of the maxillary canine. The crown height of the maxil-

lary central incisor showed the smallest size reduction. The CRI showed significant reduction only for the mandibular lateral incisor.

Both maxillary and mandibular premolars showed significantly reduced crown height and root length compared with normal controls (Table 3). Generally, the premolars demonstrated a greater percentage reduction (range, 7.6–19.7) than incisors and canines. Root length showed greater reduction than crown height, highest value being observed for the maxillary second premolars. The mandibular second premolar showed significantly reduced CRI, which indicates altered tooth proportions.

### Isochromosome and mosaic karyotypes

Table 2 presents the measurements separately for isochromosome for the long arm of X and mosaic karyotypes. Owing to the small sample sizes the testing included mosaics and isochromosomes as one group compared with controls.

The crown height was significantly reduced for maxillary incisors and the mandibular lateral incisor and canine. Root length was significantly reduced for maxillary incisors and the canine and mandibular central incisor. The maxillary central incisor had significantly reduced CRI, whereas the other incisors and canines showed normal tooth proportions.

In the isochromosome group reductions ranged from 4.6% for the crown height of the maxillary canine to 37.3% for the root length of the maxillary central incisor. The mosaic group showed reductions ranging from 3.4% to 16.1%, the greatest reduction being found for the root length of the maxillary central incisor.

Crown height and root length were significantly reduced for both mandibular and maxillary premolars (Table 3). The CRI was significantly reduced for the second mandibular premolar only. Size reduction ranged from 11.9% to 31.9% in the isochromosome and from 6.2% to 16.2% in the mosaic karyotypes. Both karyotypes showed the smallest reduction in crown height of the mandibular second premolars and largest in

Table 2. Root length and crown height of incisors and canines in 24 45X girls, 9 girls with isochromosome of the long arm of X and mosaic karyotypes, and 33 normal control girls. Mean value ( $\bar{x}$ ), standard deviation (SD). Asterisks indicate significant differences of 45X versus controls and isochromosome and mosaic karyotypes as one group versus controls. Isochromosome and mosaic karyotypes versus 45X showed no significant differences

|                         | 45X      |           |      | Isochromosomes |           |      | Mosaics  |           |      | Controls |           |      |
|-------------------------|----------|-----------|------|----------------|-----------|------|----------|-----------|------|----------|-----------|------|
|                         | <i>n</i> | $\bar{x}$ | SD   | <i>n</i>       | $\bar{x}$ | SD   | <i>n</i> | $\bar{x}$ | SD   | <i>n</i> | $\bar{x}$ | SD   |
| <b>Crown height</b>     |          |           |      |                |           |      |          |           |      |          |           |      |
| <b>Maxilla</b>          |          |           |      |                |           |      |          |           |      |          |           |      |
| I <sup>1</sup>          | 17       | 9.03*     | 0.95 | 2              | 7.95**    | 0.00 | 4        | 8.68**    | 0.89 | 33       | 9.46      | 0.71 |
| I <sup>2</sup>          | 16       | 7.96**    | 0.56 | 2              | 7.50**    | 0.14 | 3        | 7.62**    | 0.85 | 31       | 8.61      | 0.72 |
| C                       | 9        | 8.62**    | 0.75 | 2              | 9.10      | 0.14 | 2        | 8.15      | 2.19 | 21       | 9.54      | 0.66 |
| <b>Mandible</b>         |          |           |      |                |           |      |          |           |      |          |           |      |
| I <sup>1</sup>          | 16       | 7.73**    | 0.51 | 2              | 7.73      | 0.04 | 4        | 8.01      | 0.75 | 33       | 8.29      | 0.62 |
| I <sup>2</sup>          | 13       | 7.92**    | 0.66 | 2              | 7.65*     | 0.07 | 4        | 7.98*     | 0.76 | 30       | 8.59      | 0.64 |
| C                       | 9        | 8.47**    | 0.48 | 2              | 8.60*     | 0.00 | 2        | 7.73*     | 1.31 | 24       | 9.16      | 0.53 |
| <b>Root length</b>      |          |           |      |                |           |      |          |           |      |          |           |      |
| <b>Maxilla</b>          |          |           |      |                |           |      |          |           |      |          |           |      |
| I <sup>1</sup>          | 17       | 13.67***  | 2.46 | 2              | 9.95**    | 1.34 | 4        | 13.33**   | 1.49 | 33       | 15.88     | 2.00 |
| I <sup>2</sup>          | 16       | 13.59**   | 2.34 | 2              | 11.63**   | 2.30 | 3        | 13.02**   | 1.34 | 31       | 15.48     | 1.30 |
| C                       | 9        | 15.36**   | 1.59 | 2              | 12.68*    | 0.88 | 2        | 15.55*    | 1.48 | 21       | 17.86     | 2.39 |
| <b>Mandible</b>         |          |           |      |                |           |      |          |           |      |          |           |      |
| I <sup>1</sup>          | 16       | 12.44**   | 1.88 | 2              | 11.23*    | 1.24 | 4        | 12.71*    | 1.97 | 33       | 14.05     | 1.41 |
| I <sup>2</sup>          | 13       | 13.68***  | 1.48 | 2              | 14.00     | 4.03 | 4        | 14.53     | 0.29 | 30       | 15.83     | 1.59 |
| C                       | 9        | 16.24     | 1.43 | 2              | 17.07     | 1.52 | 2        | 15.95     | 0.07 | 24       | 17.37     | 1.56 |
| <b>Crown-root index</b> |          |           |      |                |           |      |          |           |      |          |           |      |
| <b>Maxilla</b>          |          |           |      |                |           |      |          |           |      |          |           |      |
| I <sup>1</sup>          | 17       | 1.53      | 0.32 | 2              | 1.25*     | 0.17 | 4        | 1.55*     | 0.20 | 33       | 1.68      | 0.22 |
| I <sup>2</sup>          | 16       | 1.72      | 0.35 | 2              | 1.55      | 0.34 | 3        | 1.71      | 0.08 | 31       | 1.81      | 0.18 |
| C                       | 9        | 1.79      | 0.20 | 2              | 1.39      | 0.12 | 2        | 1.95      | 0.34 | 21       | 1.89      | 0.31 |
| <b>Mandible</b>         |          |           |      |                |           |      |          |           |      |          |           |      |
| I <sup>1</sup>          | 16       | 1.61      | 0.24 | 2              | 1.45      | 0.17 | 4        | 1.60      | 0.30 | 33       | 1.70      | 0.18 |
| I <sup>2</sup>          | 13       | 1.73*     | 0.17 | 2              | 1.83      | 0.54 | 4        | 1.83      | 0.19 | 30       | 1.85      | 1.85 |
| C                       | 9        | 1.92      | 0.18 | 2              | 1.99      | 0.18 | 2        | 2.09      | 0.35 | 24       | 1.90      | 1.86 |

\* $p < 0.05$ ; \*\* $p < 0.01$ ; \*\*\* $p < 0.001$ .

root length of the second maxillary premolar.

No significant differences with regard to crown height, root length, and CRI were found by comparing 45X, isochromosome, and mosaic groups.

#### Root morphology of maxillary teeth

Apart from being shorter and more obtuse, incisors and canines did not differ from the controls (Fig. 2).

First premolars with two or three root components (Fig. 3) were found significantly more often in the Turner group than in controls (Table 4). Second premolars showed no differences in number of root components. In 45X patients no first pre-

molars had non-separate roots, 11 were two-rooted, and 2 were three-rooted. The sample sizes were small, so statistical testing between the karyotypes could not be performed.

Maxillary molars were three-rooted with two buccal and one palatal root component in both Turner and control patients.

#### Root morphology of mandibular teeth

Some central incisors had very short roots with a rounded apex. Otherwise, the root morphology of incisors and canines was similar to that found in the control group.

Premolars showed a complex root morphology, which was characterized by great variation and a high prevalence of root di-

Table 3. Root length and crown height of premolars in 24 45X girls, 9 girls with isochromosome of the long arm of X and mosaic karyotypes, and 33 normal control girls. Mean value ( $\bar{x}$ ), standard deviation (SD). Asterisks indicate significant differences of 45X versus controls and isochromosome and mosaic karyotypes as one group versus controls. Isochromosome and mosaic karyotypes versus 45X showed no significant differences

|                  | 45X      |           |      | Isochromosomes |           |      | Mosaics  |           |      | Controls |           |      |
|------------------|----------|-----------|------|----------------|-----------|------|----------|-----------|------|----------|-----------|------|
|                  | <i>n</i> | $\bar{x}$ | SD   | <i>n</i>       | $\bar{x}$ | SD   | <i>n</i> | $\bar{x}$ | SD   | <i>n</i> | $\bar{x}$ | SD   |
| Crown height     |          |           |      |                |           |      |          |           |      |          |           |      |
| Maxilla          |          |           |      |                |           |      |          |           |      |          |           |      |
| PM <sup>1</sup>  | 9        | 8.89***   | 0.56 | 2              | 8.33**    | 0.32 | 4        | 9.18**    | 0.90 | 20       | 10.10     | 0.61 |
| PM <sup>2</sup>  | 10       | 8.21***   | 0.46 | 2              | 7.98***   | 0.04 | 4        | 8.38***   | 0.48 | 18       | 9.56      | 0.62 |
| Mandible         |          |           |      |                |           |      |          |           |      |          |           |      |
| PM <sup>1</sup>  | 13       | 7.93***   | 0.41 | 2              | 7.15**    | 0.28 | 3        | 7.73**    | 0.84 | 22       | 8.76      | 0.60 |
| PM <sup>2</sup>  | 12       | 7.76**    | 0.39 | 2              | 7.40*     | 0.14 | 3        | 7.88*     | 0.49 | 15       | 8.40      | 0.64 |
| Root length      |          |           |      |                |           |      |          |           |      |          |           |      |
| Maxilla          |          |           |      |                |           |      |          |           |      |          |           |      |
| PM <sup>1</sup>  | 9        | 15.08**   | 2.13 | 2              | 13.60*    | 1.91 | 4        | 15.50*    | 2.29 | 20       | 17.46     | 1.73 |
| PM <sup>2</sup>  | 10       | 13.57***  | 1.53 | 2              | 11.50**   | 0.07 | 4        | 14.15**   | 2.87 | 18       | 16.89     | 2.00 |
| Mandible         |          |           |      |                |           |      |          |           |      |          |           |      |
| PM <sup>1</sup>  | 13       | 16.39***  | 1.30 | 2              | 14.90**   | 2.12 | 3        | 16.38**   | 0.18 | 22       | 18.44     | 1.61 |
| PM <sup>2</sup>  | 12       | 16.51***  | 2.09 | 2              | 12.55**   | 4.60 | 3        | 16.78**   | 0.97 | 15       | 19.19     | 1.16 |
| Crown-root index |          |           |      |                |           |      |          |           |      |          |           |      |
| Maxilla          |          |           |      |                |           |      |          |           |      |          |           |      |
| PM <sup>1</sup>  | 9        | 1.70      | 0.26 | 2              | 1.63      | 0.29 | 4        | 1.68      | 0.11 | 20       | 1.73      | 0.20 |
| PM <sup>2</sup>  | 10       | 1.65      | 0.18 | 2              | 1.44      | 0.02 | 4        | 1.68      | 0.29 | 18       | 1.77      | 0.22 |
| Mandible         |          |           |      |                |           |      |          |           |      |          |           |      |
| PM <sup>1</sup>  | 13       | 2.07      | 0.17 | 2              | 2.08      | 0.21 | 3        | 2.13      | 0.22 | 22       | 2.10      | 0.17 |
| PM <sup>2</sup>  | 12       | 2.13*     | 0.23 | 2              | 1.69*     | 0.59 | 3        | 2.13*     | 0.03 | 15       | 2.29      | 0.15 |

\* $p < 0.05$ ; \*\* $p < 0.01$ ; \*\*\* $p < 0.001$ .

vision (Figs. 4A–F). Some of the root types, were quite unusual for this tooth type. The such as the molar-like premolar (Fig. 4D), root morphology varied within the same



Fig. 2. Short and obtuse roots of maxillary incisors in a 45X karyotype.



Fig. 3. Three-rooted maxillary first premolar in a Turner patient (45X).

Table 4. Maxillary premolars distributed on the basis of root morphology and karyotype

|                  | No. of patients | No. of teeth      |                              |                                |
|------------------|-----------------|-------------------|------------------------------|--------------------------------|
|                  |                 | Non-separate root | Two separate root components | Three separate root components |
| First premolars  |                 |                   |                              |                                |
| 45X              | 24              | 0                 | 11                           | 2                              |
| 46X, i(Xq)       | 3               | 1                 | 1                            | 0                              |
| Mosaics          | 6               | 1                 | 4                            | 0                              |
| Turner, total    | 33              | 2                 | 15                           | 2                              |
| Controls         | 33              | 11                | 20                           | 0                              |
| Second premolars |                 |                   |                              |                                |
| 45X              | 24              | 10                | 3                            | 1                              |
| 46X, i(Xq)       | 3               | 0                 | 1                            | 0                              |
| Mosaics          | 6               | 4                 | 1                            | 0                              |
| Turner, total    | 33              | 14                | 5                            | 1                              |
| Controls         | 33              | 20                | 5                            | 0                              |

Turner versus controls: first premolars, chi-square = 4.2;  $p < 0.05$ .

patient, and often corresponding teeth from the left and right side were different. In the control material 90.0% of the first and 100% of the second premolars were one-rooted, with one root canal. In the Turner group there was a significantly increased number of first premolars with root morphology different from this type (Table 5).

Mandibular molars also showed great variation in root morphology (Figs. 5A–H), and

supernumerary root components were frequently seen. In the controls 93.9% of the first molars were two-rooted, and 93.3% of the second molars were either of this type or had fused mesial and distal root. There was a significantly increased number of first molars with root morphology different from these types in the Turner material (Table 6).

The greatest variation in root morphology was found in the 45X group. Because of



Fig. 4. Root types of mandibular premolars. A) Type 1: one-root and non-separate root canal; B) type 2: one root with separate root canals; C) type 3: separate buccal and lingual root components; and D–F) type 4: separate mesial and distal root components; degree of separation, 0.6 (D), 0.5 (E), and 0.2 (F).

Table 5. Mandibular premolars distributed on the basis of root morphology and karyotype

|                  | No. of patients | No. of teeth |        |        |        |
|------------------|-----------------|--------------|--------|--------|--------|
|                  |                 | Type 1       | Type 2 | Type 3 | Type 4 |
| First premolars  |                 |              |        |        |        |
| 45X              | 24              | 2            | 3      | 8      | 2      |
| 46X,i(Xq)        | 3               | 0            | 0      | 2      | 0      |
| Mosaics          | 6               | 2            | 1      | 2      | 0      |
| Turner, total    | 33              | 4            | 4      | 12     | 2      |
| Controls         | 33              | 27           | 2      | 1      | 0      |
| Second premolars |                 |              |        |        |        |
| 45X              | 24              | 7            | 2      | 1      | 4      |
| 46X,i(Xq)        | 3               | 1            | 0      | 0      | 1      |
| Mosaics          | 6               | 4            | 0      | 1      | 0      |
| Turner, total    | 33              | 12           | 2      | 2      | 5      |
| Controls         | 33              | 25           | 0      | 0      | 0      |

Turner versus controls: first premolars, chi-square = 27.2;  $p < 0.001$ .

45X versus controls: first premolars, chi-square = 25.7;  $p < 0.001$ .

the small sample size statistical comparisons between the karyotypes could not be performed.

#### Taurodontism

In a 45X patient a second mandibular premolar was classified as hypertaurodont (Fig. 4F). The tooth had little constriction at the cervix and a broad prism-shaped root with about the same thickness from the cervix to the apex and an enlarged pulp cavity with apical bifurcation. Taurodont molars were not found in Turner or control patients.

#### Discussion

In the investigated Turner patients we found reduced root length and crown height of incisors, canines, and premolars. Some of these teeth also showed reduced CRI, indicating altered tooth proportions. A more complex root morphology of mandibular premolars and molars was also seen. Our findings, which are in agreement with those of Filipsson et al. (18), indicate that short roots are part of the Turner syndrome. Another investigation describes the presence of a large number of separate roots in mandibular premolars (19), which is also a finding in our investigation and confirms the influence of X-chromosome deficiency on

root formation. The high prevalence of supernumerary roots of mandibular molars has not been reported earlier.

Study of root length and morphology in vivo requires indirect methods, and radiographs are regularly used. There are some limitations in using this method, especially in morphologic studies. An X-ray image has only two dimensions, whereas the original structure has three. Even though the combined viewing of two or more different projections provides information about the three-dimensional composition of an object, problems with regard to masking of structures often remain. Distortion problems associated with intraoral radiography are partly overcome by use of the paralleling technique (20). In the premolar regions we decided to measure root length from panoramic radiographs, since this technique gives good reproducibility of the vertical dimension in the lateral segments (27), and distortion associated with intraoral film placement is avoided, which is important in a group of patients with a narrow maxillary arch (7, 8).

Studies of tooth crown size and morphology in Turner patients have shown reduction in both mesiodistal and buccolingual dimensions. These morphologic alterations are found for all types of teeth and are caused by reduced thickness of the enamel layer (16). Reduction in cusp number and size is a



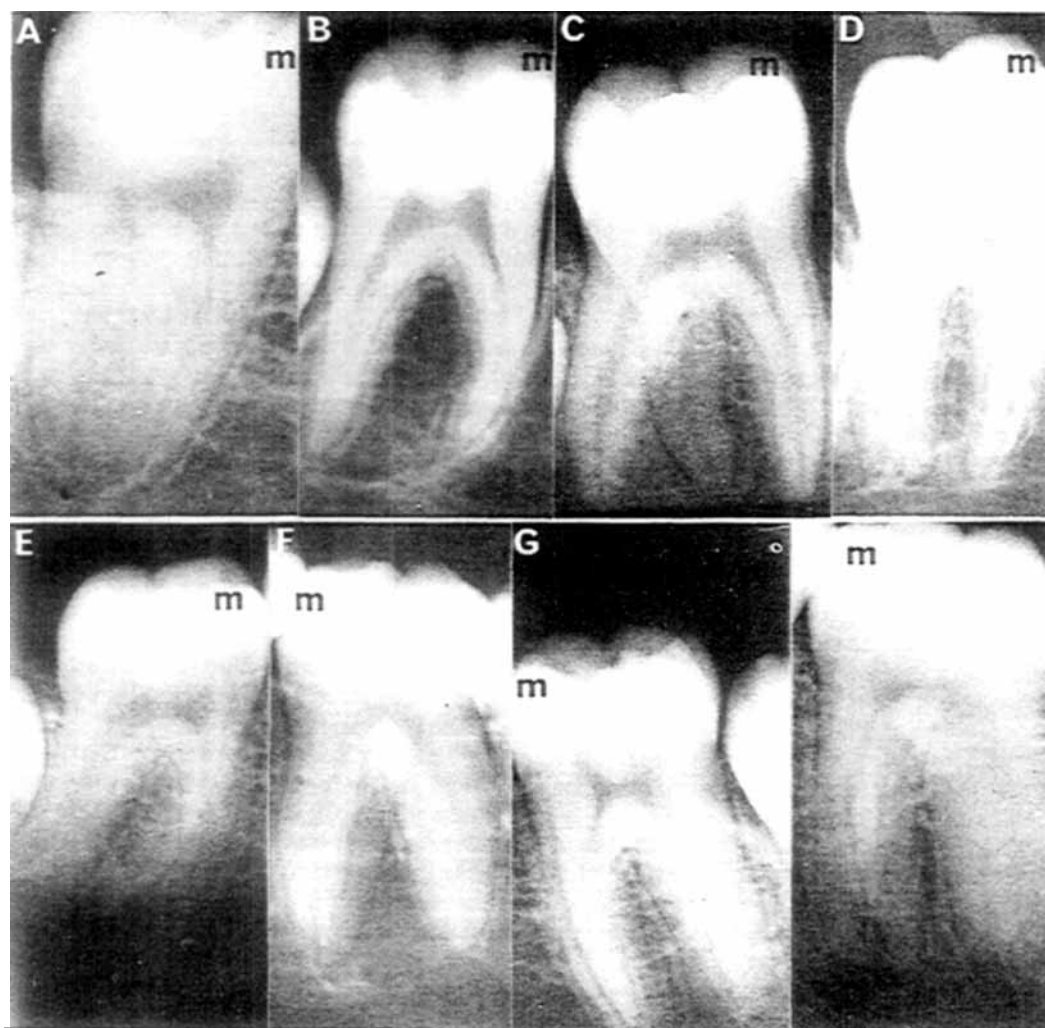


Fig. 5. Root types of mandibular molars. The letter *m* indicates the mesial side. A) Type 1: fused mesial and distal root components; B) type 2: separate mesial and distal root components; C) type 3: separate mesial, distal, and distolingual root components (distolingual root identified as radix entomolaris); D) type 4: separate mesial and two separate distal root components; E) type 5: two separate mesial and one separate distal root components; and type 6: molar with separate mesial and distal root components, of which either the mesial (F), distal (G), or both (H) root components show abnormal width and separate apices.

characteristic, as are reduced prevalence of Carabelli's trait and shovel shape (15, 17). In sum, the morphologic alterations give a simplified crown morphology, which is in contrast to our findings of a complex root morphology. Even though the findings differ for crown and root morphology, it seems probable that there is an interrelationship between the abnormal features observed.

Disturbed or restricted growth is the most predominant characteristic of girls with the Turner syndrome, besides various somatic abnormalities (28–31). Lippe (3) has suggested a hypothesis to explain most of the anomalies observed in Turner syndrome patients. In her opinion the basic defect is growth disorder of the mesenchymal tissue, which leads to skeletal and vascular growth

Table 6. Mandibular molars distributed on the basis of root morphology and karyotype

|               | No. of patients | No. of teeth |        |        |        |        |        |
|---------------|-----------------|--------------|--------|--------|--------|--------|--------|
|               |                 | Type 1       | Type 2 | Type 3 | Type 4 | Type 5 | Type 6 |
| First molars  |                 |              |        |        |        |        |        |
| 45X           | 24              | 0            | 16     | 2      | 2      | 1      | 3      |
| 46X,i(Xq)     | 3               | 0            | 1      | 0      | 0      | 1      | 1      |
| Mosaics       | 6               | 0            | 4      | 1      | 1      | 0      | 0      |
| Turner, total | 33              | 0            | 21     | 3      | 3      | 2      | 4      |
| Controls      | 33              | 0            | 31     | 0      | 0      | 0      | 2      |
| Second molars |                 |              |        |        |        |        |        |
| 45X           | 24              | 0            | 5      | 0      | 0      | 3      | 1      |
| 46X,i(Xq)     | 3               | 0            | 1      | 0      | 0      | 0      | 1      |
| Mosaics       | 6               | 1            | 2      | 0      | 0      | 0      | 2      |
| Turner, total | 33              | 1            | 8      | 0      | 0      | 3      | 4      |
| Controls      | 33              | 2            | 12     | 0      | 0      | 0      | 1      |

Turner versus controls: first molars, chi-square = 9.5;  $p < 0.01$ .

abnormalities. The significance of this mechanism for tooth crown morphology has previously been discussed (17). There is also evidence that the dental papilla, which is of mesenchymal origin, influences early root formation and determines whether the enamel organ will form an incisor or a molar tooth (32). Experiments have shown that if the dental papilla of a molar tooth germ is combined with the dental enamel organ of an incisor tooth germ, the dental enamel organ assumes the shape of the molar. Likewise, incisor dental papilla can cause a molar dental organ to become an incisor (33). These experiments indicate that the ectomesenchyme not only induces the development of a tooth but also determines its shape.

Alexandersen & Carlsen (34) state that root formation is based on the differential growth of the dental papilla within the presence of Hertwig's epithelial root sheet. Interactions between epithelium and connective mesenchymal tissue are involved in many aspects of dental development, such as the determination of tooth crown pattern and the shape, size, and numbers of roots (33). The mechanisms by which this occurs are, however, unknown.

Some studies of dental size and morphology in persons with sex-chromosome

aneuploidy conclude that genes of the X chromosome directly influence both enamel and root formation (16, 35–37). In our opinion the alterations in dental size and morphology observed indicate that the interactions between mesenchyme and epithelium are in some way influenced by the reduced amount of X-chromosome material, thereby modifying the whole sequence of events in tooth formation. Several other mechanisms have been proposed to explain the increased variance of morphometric traits in aneuploidy conditions (38–41).

Alteration in cellular activity of tooth germs and decreased developmental stability are the theories most often discussed. Townsend & Brown (39) and Bell et al. (46) have pointed out that the theories of altered cellular activity and decreased developmental stability need not be considered mutually exclusive but rather as consequences of each other.

The roots were significantly shorter both in the maxilla and in the mandible, but the morphologic alterations were mainly found in the mandibular premolars and molars. In contrast, traits or irregularities in crown morphology were most prevalent in the maxilla (17). In a Danish material of normal individuals the prevalence of mandibular premolars with two separate root com-

ponents was reported to be 10% and 5% of first and second premolars, respectively (22). In the Turner patients the frequency of premolars with separate roots was increased. Especially the morphologic type with separate mesial and distal root components, type 4 (Fig. 4DE), seems to be specific for Turner syndrome patients. Both the unusually large degree of separation and the divergence between the mesial and the distal root components make the tooth molariform. Such mandibular premolars are rarely seen in the normal population (22).

In European populations separate radix entomolaris is reported to be present on mandibular first molars with a maximum frequency of 3.4%, whereas the corresponding maximum frequency in Mongolian populations is 43.7% (42). The high frequency of this supernumerary root structure in certain Mongolian populations indicates a heritable basis for the trait. In the Turner patients a supernumerary radix entomolaris was often present on first mandibular molars. In addition, three first molars were three-rooted with a mesial and two separate distal root components, the lingual of which might be a radix entomolaris.

In the 45X karyotype we also observed three-rooted first and second molars with two separate mesial and one separate distal root component. The mesial supernumerary root could be a radix paramolaris, which occurs in low frequencies in mandibular molars (22). Several molars from all three karyotypes had greater width of the mesial, distal, or both root components. Together with the number of apices this indicated the presence of supernumerary root structures. From our observations it seems that the tendency for increased root division in mandibular molars is associated with all karyotypes of the syndrome.

Chromosomal aneuploidy is known to influence pulp and root morphology by increasing the incidence of taurodontism (35, 43). The trait has mainly been associated with an additional number of X chromosomes, whereas the prevalence in patients with X-chromosome monosomy is reported to be normal (44). This is supported by our investigation, in which no molars and only

one premolar showed this trait. Taurodontism is reported to be part of several syndromes (36, 37, 41, 43, 45, 46), and increased incidence of the trait has been associated with those having an ectodermal defect (43, 45, 47).

The development of teeth involves many complex biologic processes, such as epithelial-mesenchymal relationships, morphogenesis, fibrillogenesis, and mineralization. The dental anomalies observed in Turner syndrome indicate that several of these processes are influenced by the X-chromosome deficiency.

*Acknowledgements.*—Appreciation is expressed to the Department of Pediatrics, University of Bergen, for their initiative to collaborate with the Departments of Orthodontics and Oral Radiology in this field.

## References

1. Nielsen J, Wohler M. Sex chromosome abnormalities found among 34,910 newborn children: result from a 13-year incidence study in Århus, Denmark. *Birth Defects* 1991;26:209-23.
2. Lemli L, Smith DW. The XO syndrome: a study of the differentiated phenotype in 25 patients. *J Pediatrics* 1963;63:577-88.
3. Lippe BM. Physical and anatomical abnormalities in Turner syndrome. In: Rosenfeld RG, Grumbach MM, editors. *Turner syndrome*. New York: Marcel Dekker Inc, 1990.
4. Connor JM, Ferguson-Smith MA. *Essential medical genetics*. 3rd ed. Oxford: Blackwell Scientific Publications, 1991.
5. Grumbach MM, Conte FA. Disorders of sex differentiation. In: Wilson JD, Foster DW, editors. *Textbook of endocrinology*. Philadelphia (PA): W.B. Saunders Company, 1992:884-99.
6. Jensen BL. Craniofacial morphology in Turner syndrome. *J Craniofac Genet Dev Biol* 1985;5:327-40.
7. Laine T, Alvesalo L, Lammi S. Palatal dimension in 45,X-females. *J Craniofac Genet Dev Biol* 1985;5:239-46.
8. Laine T, Alvesalo L. Size of the alveolar arch of the mandible in relation to that of the maxilla in 45,X females. *J Dent Res* 1986;65:1432-4.
9. Laine T, Alvesalo L, Savolainen A, Lammi S. Occlusal morphology in 45,X females. *J Craniofac Genet Dev Biol* 1986;6:351-5.
10. Harju M, Laine T, Alvesalo L. Occlusal anomalies in 45,X/46,XX- and 46,Xi(Xq)-women (Turner syndrome). *Scand J Dent Res* 1989;97:387-91.
11. Townsend C, Jensen BL, Alvesalo L. Reduced tooth size in 45X (Turner syndrome) females. *Am J Phys Anthropol* 1984;65:367-71.
12. Townsend G, Alvesalo L, Jensen BL, Kari M. Patterns of tooth size in human chromoso-

- mal aneuploidies. In: Russell DE, Santoro J-P, Sigogneau-Russell D, editors. Teeth revisited. Proceedings of the VIIth International Symposium on Dental Morphology. Mus Nat Hist C:53. Paris: Museum of Natural History, 1988:25-45.
13. Mayhall J, Alvesalo L, Townsend G. Tooth crown size in 46,X,i(XQ) females (abstract). *J Dent Res* 1987;66(Spec Iss): abstract 22.
  14. Varrela J, Townsend G, Alvesalo L. Tooth crown size in human females with 45,X/46,XX chromosomes. *Arch Oral Biol* 1988;33:291-4.
  15. Kirveskari P, Alvesalo L. Dental morphology in Turner's syndrome (45,X females). In: Kurten B, editor. Teeth: form, function and evolution. New York: Columbia University Press, 1982:298-303.
  16. Alvesalo L, Tammisalo E. Enamel thickness in 45,X females' permanent teeth. *Am J Hum Genet* 1981;33:464-9.
  17. Midtbø M, Halse A. Tooth crown size and morphology in Turner syndrome. *Acta Odontol Scand* 1994;52:7-19.
  18. Filipsson R, Lindsten J, Almquist S. Time of eruption of the permanent teeth, cephalometric and tooth measurements and sulphation factor activity in 45 patient with Turner syndrome with different types of chromosome aberrations. *Acta Endocrinol* 1965;48:91-113.
  19. Varrela J. Root morphology of mandibular premolars in human 45,X females. *Arch Oral Biol* 1990;35:109-12.
  20. Larheim TA, Eggen S. Determination of tooth length with a standardized paralleling technique and calibrated radiographic measuring film. *Oral Surg Oral Med Oral Pathol* 1979;48:374-8.
  21. Thanyakarn C, Hansen K, Rohlin M, Åkesson L. Measurements of tooth length in panoramic radiographs. I. The use of indicators. *Dentomaxillofac Radiol* 1992;21:26-30.
  22. Carlsen O. Dental morphology. Copenhagen: Munksgaard, 1987.
  23. Shaw JCM. Taurodont teeth in South African races. *J Anat* 1928;62:476-98.
  24. Madeira MC, Leite HF, Niccoli Filho WD, Simões A. Prevalence of taurodontism in premolars. *Oral Surg Oral Med Oral Pathol* 1986;61:158-62.
  25. Dahlberg G. Statistical methods for medical and biological students. London: Allen and Unwin, 1940.
  26. Ryan BF, Joiner BL, Ryan TA. Minitab handbook. 2nd ed. Boston: Duxbury Press, 1985.
  27. Larheim TA, Svanaes DB, Johannessen S. Reproducibility of radiographs with the Orthopantomograph 5: tooth-length assessment. *Oral Surg Oral Med Oral Pathol* 1984;58:736-41.
  28. Lindsten J, Filipsson R, Hall K, Leikrans S, Gustavson K-H, Ryman N. Body height and dental development in patients with Turner's syndrome. *Helv Paediat Acta* 1974;34 Suppl:33-46.
  29. Park E. Body shape in Turner's syndrome. *Hum Biol* 1977;49:215-23.
  30. Park E, Bailey JD, Cowell CA. Growth and maturation of patients with Turner's syndrome. *Pediatr Res* 1983;17:1-7.
  31. Pelz L, Timm D, Eyermann E, Hinkel GK, Kirchner M, Verron G. Body height in Turner's syndrome. *Clin Genet* 1982;22:62-6.
  32. Bhaskar SN editor. Orban's oral histology and embryology. 10th ed. St Louis: C.V. Mosby Company, 1986:40-4, 169.
  33. Ten Cate AR. Epithelial and mesenchymal relations. In: Ten Cate AR, editor. Oral histology: development, structure, and function. St Louis (MO): C.V. Mosby Company, 1985.
  34. Alexandersen V, Carlsen O. Underkæbevisdomstandens rodkompleks. II. En morfogenetisk vurdering. *Tandlaegebladet* 1985;89:353-62.
  35. Varrela J, Alvesalo L. Taurodontism in females with extra X chromosomes. *J Craniofac Genet Dev Biol* 1989;9:129-33.
  36. Varrela J, Alvesalo L. Taurodontism in 47,XXY males: an effect of the extra X chromosome on root development. *J Dent Res* 1988;67:501-2.
  37. Alvesalo L, Varela J. Taurodontism and the presence of an extra Y chromosome: study of 47,XXY males and analytical review. *Hum Biol* 1991;63:31-8.
  38. Barlow P. The influence of inactive chromosomes on human development. *Human Genet* 1973;17:105-36.
  39. Townsend GC, Brown RH. Dental crown variant in children and young adults with Down syndrome. *Acta Odontol Pediat* 1986;7:35-9.
  40. Shapiro BL. Down syndrome. A disruption of homeostasis. *Am J Med Genet* 1983;14:241-69.
  41. Witkop CJ Jr, Keenan KM, Cervenka J, Jaspers MT. Taurodontism: an anomaly of teeth reflecting disruptive developmental homeostasis. *Am J Med Genet* 1988;4 Suppl:85-97.
  42. Carlsen O, Alexandersen V. Radix entomolaris: identification and morphology. *Scand J Dent Res* 1990;98:363-73.
  43. Jaspers MT, Witkop CJ Jr. Taurodontism. an isolated trait associated with syndromes and X-chromosome aneuploidy. *Am J Hum Genet* 1980;32:396-413.
  44. Varrela J, Alvesalo L, Mayhall J. Taurodontism in 45,X females. *J Dent Res* 1990;69:494-5.
  45. Crawford PJ, Aldred MJ, Clarke A. Clinical and radiographic dental findings in X linked hypohidrotic ectodermal dysplasia. *J Med Genet* 1991;28:181-5.
  46. Bell J, Civil CR, Townsend GC, Brown RH. The prevalence of taurodontism in Down's syndrome. *J Ment Defic Res* 1989;33:467-76.
  47. Jorgenson RJ. The condition manifesting taurodontism. *Am J Med Genet* 1982;11:435-42.

Received for publication 29 November 1993

Accepted 8 April 1994