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**Case Report**

# Morphological Study on Quadruplets by Cephalometric and Model Analyses

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## Abstract

Clarifying the genetic factors involved in maxillofacial growth and development is very important in orthodontic treatment planning and prognosis. However, few dental studies have examined multiple births. The present orthodontic evaluation was conducted using orthodontic data from a set of quadruplets. Orthodontic evaluation was performed on a set of quadruplets (1 girl and 3 boys) aged 9 years and 7 months at the initial visit. Although all 4 children weighed only about 1,400 g each at birth, height and body weight subsequently normalized. Mean skeletal age of the quadruplets was 10 years and 2 months, about 6 months ahead of their calendar age. In all 4 children, facial profile was mostly symmetrical and convex. Intraoral findings showed a Hellman's dental age of IIIA, together with spacing of the upper anterior teeth. Both overbite and overjet were 5–7 mm, and mesial step of the terminal plane was noted. Model analysis showed that tooth materials were on the large side, while arch width was narrow. Cephalometric analysis revealed that the ANB of the first- and fourth-born children was 6°, and skeletal maxillary protrusion due to mandibular retrusion was diagnosed. The second- and third-born children exhibited no marked skeletal abnormalities.

Key words: Multiple fetuses—Quadruplets—Orthodontic treatment—Cephalometric analysis—Model analysis

## Introduction

Before 1990, the incidence of multiple-fetus pregnancy involving ≥4 fetuses accounted for only several cases per million deliveries, but due to advances in assisted-reproduction technology, the incidence of multiple-fetus pregnancy rapidly increased to 38 cases per million deliveries in 1994 and 34 cases per million deliveries in 1995. However, the incidence of multiple-fetus pregnancy has decreased markedly since 1996. The various medical, social, economic and family issues facing parents with multiple-fetus pregnancy have raised caution on the use of fertility drugs and reduced fetus-reduction surgery[12,15].

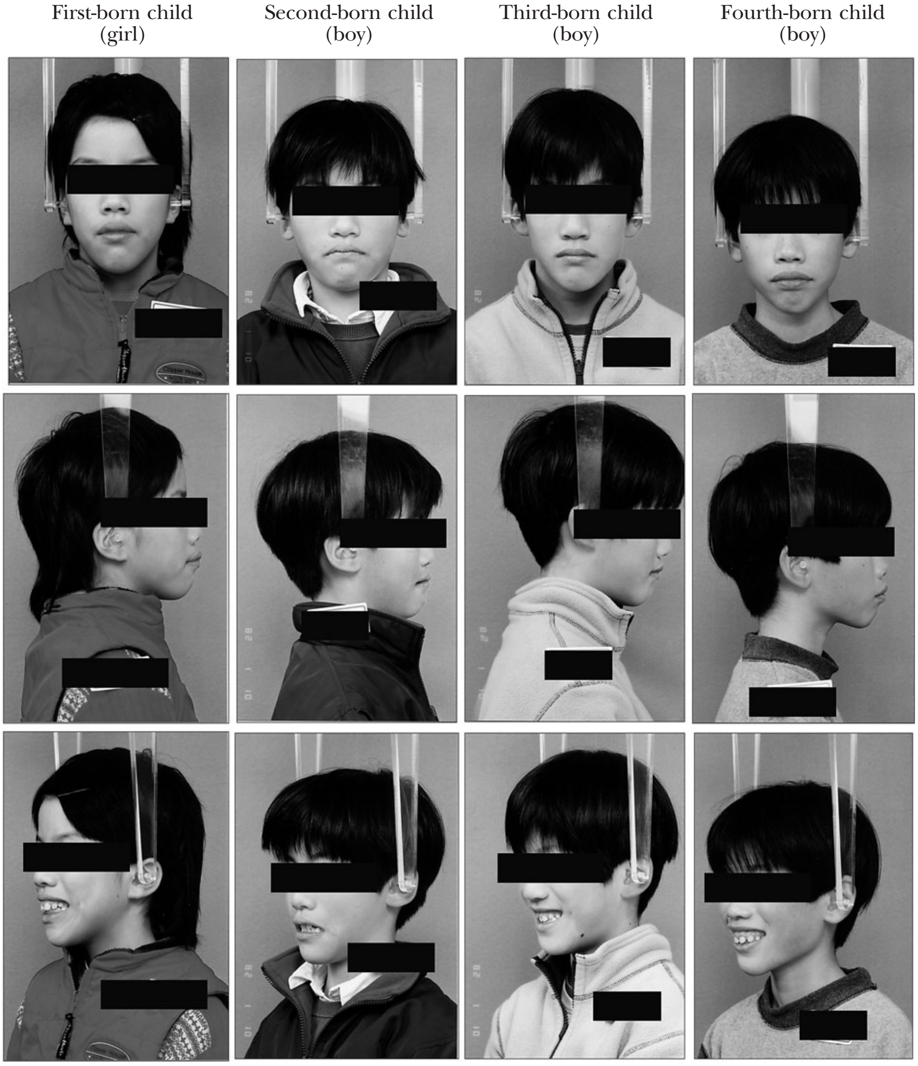


Fig. 1 Facial photographs at age 9y7m

Clarifying the genetic factors involved in maxillofacial growth and development is very important in orthodontic treatment planning and prognosis[11)]. Therefore, for a long time, studies of twins have been conducted to analyze congenital abnormalities and biological activities[6)]. In dentistry, studies have been conducted to ascertain maxillofacial growth and development[9)], morphological features of the palate[7)], location of congenital missing teeth[8)], location of supernumerary teeth, and similarities in tooth morphology[13)]. However, few dental studies have examined multiple births. Plaster models were used to ascertain tooth morphology, tooth width, dental arch form, and dental caries in 6-year-old quintuplets in 1981[10)] and 3-year-old quintuplets in 1989[1)]. To the best of our knowledge, no studies have compared and investigated maxillofacial morphology using cephalometric radiograph.

The present orthodontic evaluation was conducted using orthodontic data from a set of quadruplets.

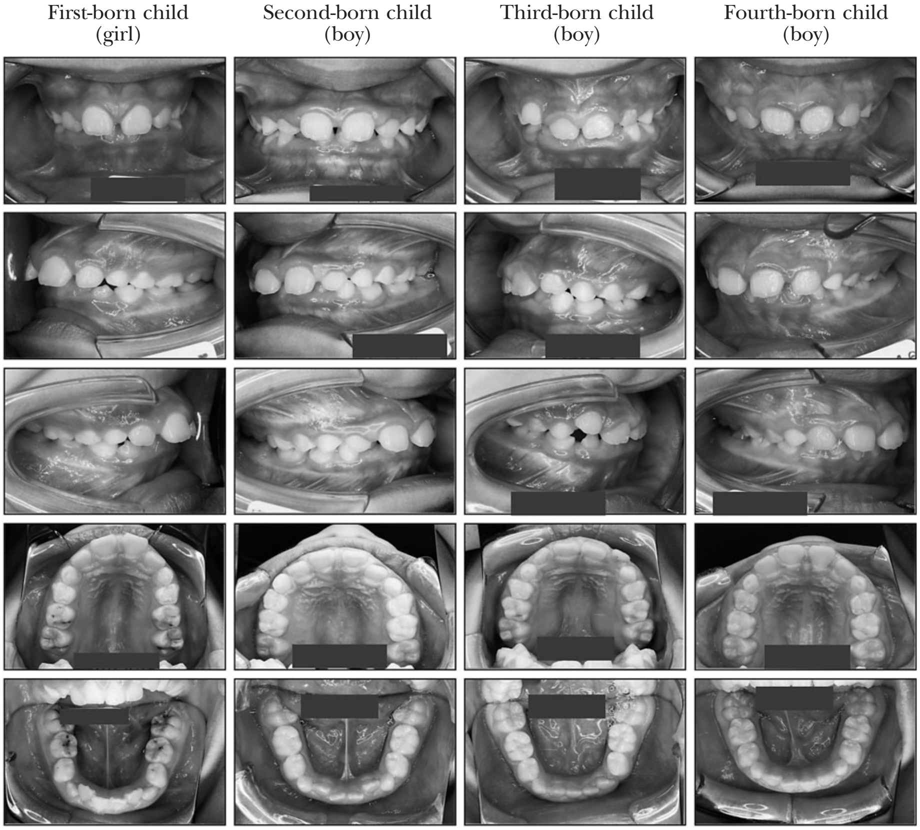


Fig. 2 Oral photographs at age 9y7m

## Case Report

The age of the quadruplets at the initial visit was 9 years and 7 months. The first-born child (girl) and the fourth-born child (boy) displayed maxillary protraction and gummy smile, and the second- and third-born children exhibited deep overbite. No systemic abnormalities were identified and nutritional status was favorable.

### 1. Facial findings

Each of the 4 children displayed a long head, and facial features were mostly symmetrical. Facial profile was convex type. In relation to the E-line, degree of lower lip protrusion was 7.9 mm for the first-born child, 3.6 mm for the second-born child, 1.8 mm for the third-born child and 7.9 mm for the fourth-born child. The first- and fourth-born children displayed marked gummy smiles, and when the lips were closed, the chin became tense (Fig. 1).

### 2. Oral findings

In all 4 children, Hellman's dental age was IIIA, and spacing of the upper anterior teeth was observed. Overbite and overjet were 5 and 10 mm for the first-born child, 4 and 5 mm for the second-born child, 4 and 5 mm for the third-born child, and 5 and 8 mm for the fourth-born child, respectively. In addition, in all 4 children, mesial-step occlusion of the second deciduous molars was noted. Median maxillary and mandibular central incisors matched the facial midline. A Carabelli cusp was seen in the first right molar of the fourth-born child (Fig. 2).

Table 1 Model analysis for the quadruplets

| | | Mean ± SD | First-born child | Second-born child | Third-born child | Fourth-born child |
|---|---|---|---|---|---|---|
| Maxillary arch | Coronal arch Anterior width | 36.9 ± 1.6 | 34.5 | 39.9 | 30.0 | 38.5 |
| | Coronal arch Posterior width | 48.3 ± 2.3 | 40.0 | 46.9 | 45.0 | 45.5 |
| | Basal arch Anterior width | 41.8 ± 2.6 | 34.0 | 46.5 | 35.0 | 46.5 |
| | Basal arch Posterior width | 62.4 ± 2.4 | 52.0 | 59.1 | 61.0 | 64.5 |
| Mandibular arch | Coronal arch Anterior width | 29.8 ± 1.1 | 26.5 | 29.9 | 25.2 | 29.0 |
| | Coronal arch Posterior width | 42.5 ± 2.3 | 35.1 | 41.2 | 39.0 | 39.4 |
| | Basal arch Anterior width | 32.6 ± 2.4 | 27.0 | 36.6 | 25.0 | 35.7 |
| | Basal arch Posterior width | 58.1 ± 2.8 | 50.0 | 59.4 | 60.0 | 58.2 |

**3. Model analysis**

In all 4 children, tooth width for the four maxillary and mandibular anterior teeth was on the large side. In terms of dental arch width, maxillary and mandibular inter-canine widths for the first- and third-born children were smaller (≥1 SD), and inter-molar width for all 4 children was small (Table 1).

**4. Cephalometric findings**

SNB for the first- and fourth-born children was 73° and 76°, respectively, and ANB was 5° and 6°, respectively. These 2 children displayed skeletal maxillary protrusion accompanied by mandibular retrusion. The second- and third-born children showed no marked abnormalities in terms of maxillomandibular relationships or incisor inclination (Fig. 3, Table 2).

**5. Physical growth curves**

The quadruplets were delivered by caesarean section. At the time of delivery, height of the first-, second-, third- and fourth-born children was 39, 38, 40, and 41 cm, respectively, and weight was 1,390, 1,130, 1,350, and 1,570 g, respectively. Current height of the first-, second-, third- and fourth-born children was 133, 122, 130, and 131 cm, respectively, and weight was 2,500, 1,980, 2,430, and 2,450 g, respectively. Compared to children in the same age group, the quadruplets had caught up in terms of height and body weight. Skeletal age of the first-, second-, third-, and fourth-born children was 10 years and 2 months, 10 years, 10 years and 2 months, and 10 years and 11 months, respectively. As a result, skeletal age of the quadruplets was about 6 months ahead of calendar age (9 years and 7 months) (Figs. 4, 5).

## Discussion

While genetic and environmental factors play a large role in malocclusion, differentiating between them is very difficult[6,11]. However, by preventing and managing caries and pernicious habits, acquired factors (environmental factors) can be equalized, and comparison and investigation of maxillofacial and cranial morphologies at the level of genetic factors may be possible in future. Twin

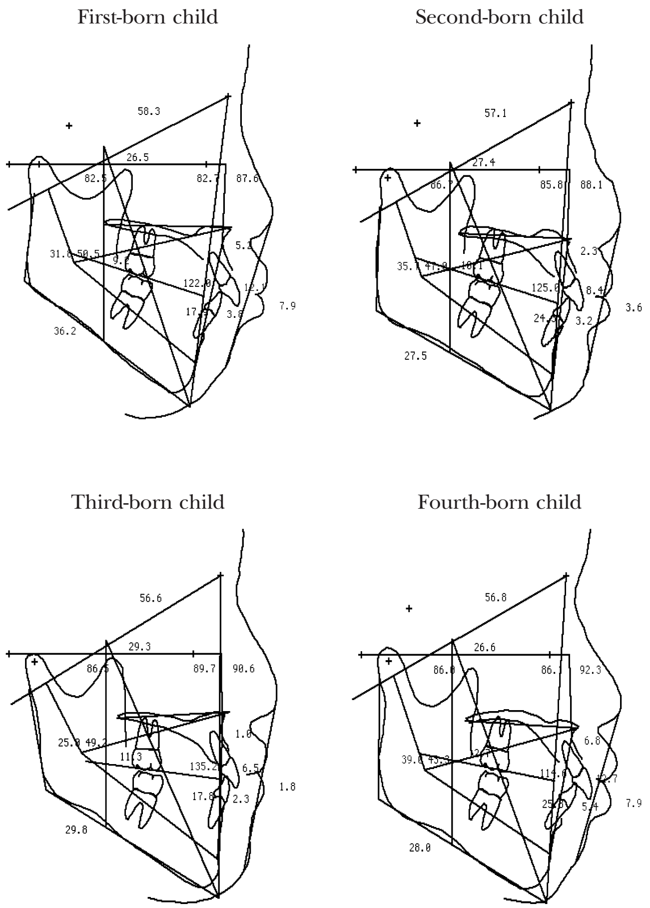


Fig. 3 Cephalometric tracing for the quadruplets (Ricketts Analysis)

Table 2 Cephalometric measurements

| | Mean ± SD | First-born child | Second-born child | Third-born child | Fourth-born child |
|---|---|---|---|---|---|
| SNA (°) | 83.2 ± 2.61 | 78.0 | 81.0 | 80.0 | 82.0 |
| SNB (°) | 80.4 ± 2.5 | 73.0 | 78.5 | 79.0 | 76.0 |
| ANB (°) | 2.8 ± 1.7 | 5.0 | 2.5 | 1.0 | 6.0 |
| FMA (°) | 24.8 ± 5.9 | 35.0 | 26.0 | 30.0 | 28.0 |
| IMPA (°) | 97.1 ± 4.9 | 86.0 | 95.0 | 80.0 | 99.0 |
| FMIA (°) | 61.6 ± 5.8 | 60.0 | 59.0 | 70.0 | 53.0 |
| Gonial angle (°) | 117.5 ± 8.1 | 121.0 | 113.0 | 135.0 | 118.0 |
| Ramus angle (°) | 88.5 ± 4.5 | 93.0 | 93.0 | 80.0 | 89.5 |
| U1-FH (°) | 110.8 ± 5.6 | 116.0 | 115.0 | 115.0 | 120.0 |
| Overjet (mm) | 2.5 | 10.0 | 6.0 | 5.0 | 8.0 |
| Overbite (mm) | 2.5 | 5.0 | 4.0 | 4.0 | 5.0 |

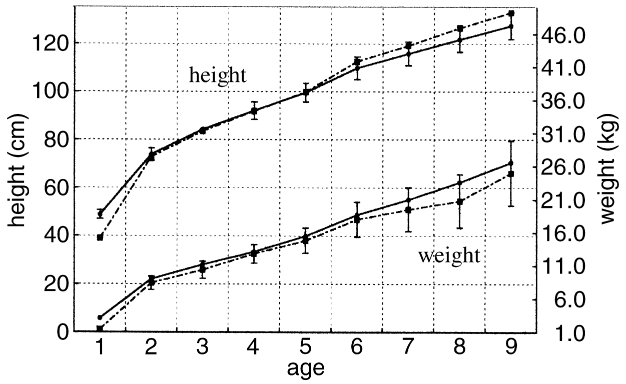


Fig. 4 Physical growth curves in girl (height, weight)
■: First-born child (girl)
●: mean and SD

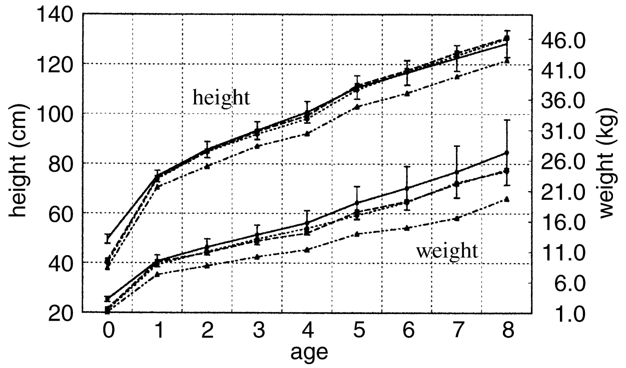


Fig. 5 Physical growth curves in boy (height, weight)
◆: Second-born child (boy)
▲: Third-born child (boy)
■: Fourth-born child (boy)
●: mean and SD

and multiple-fetus studies involving monozygotic and polyzygotic siblings are therefore very important for orthodontic research.

Model analysis showed similarities between the first- and third-born children and between the second- and fourth-born children. Also, for the quadruplets, tooth width for the 4 anterior teeth and first molar tended to be high. Miyagawa *et al.*[1] studied a set of quintuplets and reported that 40% of anterior teeth were significantly smaller than the norm for age-matched Japanese ($\geq$1 SD) and tended to be very small. Although the quadruplets in the present study were low-birth-weight infants, teeth tended to be large because of favorable maternal conditions. As far as dental arch width is concerned, Ishikawa *et al.*[7] conducted a twin study and noted that genetic factors played a large role in posterior palatal width and volume, but not in long diameter or depth. In the present study, inter-molar width of the quadruplets was narrow, but inter-canine width was narrow for only the first- and third-born children. Inter-molar width is thus genetically more stable. This may have been related to the increase in interdeciduous canine width due to eruption of lateral incisors and low tongue in the first- and third-born children. Furthermore, Miyagawa *et al.*[1] examined a set of quintuplets and suggested the possibility of zygosity diagnosis based on deciduous tooth morphology. Anders[2] also reported that zygosity diagnosis might be possible based on permanent dentition. Although few studies have compared the degree of involvement of genetic factors in phenotypic expression[14], Ishikawa *et al.*[8], Biggerstaff[5] and Aoyanagi[3] studied genetic factors for tooth morphology abnormalities such as Carabelli cusp in twins, and reported that while significant differences existed in concordance rate between mono- and polyzygotic cases, such abnormalities were not necessarily affected by genetic factors. A Carabelli cusp was seen in only one of the quadruplets, suggesting that Carabelli cusp of the first maxillary molar is not necessarily influenced by genetic factors.

Cephalometry showed skeletal maxillary protrusion for the first- and fourth-born children, but the results were mostly normal for the second- and third-born children. Unlike model analysis, skeletal patterns were comparable between the first- and fourth-born children and between the second- and third-born children. However, in terms of mandibular morphology, similarities were seen between the first- and third-born children and between the second- and fourth-born children: gonial angle of the first, second and fourth children was within 1 SD of the mean, and ramus angle of the second and fourth children was within 1 SD of the mean. Ishikawa *et al.*[9] studied Chinese female twins and reported that genetic factors accounted for 70–80% of the posteroinferior contour and size of the mandible up to early adolescence. Arya *et al.*[4] also reported that genetic factors accounted for 60–70% of mandibular development in North American Caucasian twins. Among the quadruplets investigated here, the first- and third-born children displayed different occlusions, but shared similar features based on model analysis and mandibular morphology.

All 4 children are currently undergoing orthodontic treatment, and we plan to assess the effects of early orthodontic treatment on maxillofacial growth and development in the future.

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