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ORIGINAL ARTICLE

Average craniofacial development from 6 to 35 years of age in a mixed group of patients with juvenile idiopathic arthritis

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Abstract

Objective. To investigate long term the average changes in craniofacial morphology in a group of Norwegian juvenile idiopathic arthritis (JIA) patients with mixed diagnosis from 6 to 35 years of age. A matched group of healthy individuals was included as controls. ***Material and Methods.*** Craniofacial development in 38 females and 16 males with JIA was followed on lateral cephalograms from childhood for 23–31 years. The patients were compared with the healthy individuals at the ages of 6, 9, 12, and 35 years. ***Results.*** At 6 and 9 years of age, the average craniofacial morphology in the JIA patients was similar to that of the control subjects. At 12 years of age, significant craniofacial morphological differences between the JIA and the control subjects appeared. These included a steeper mandibular plane angle and a more retrognathic position of the mandible. The length of the mandible (ar-gn), the height of the ramus (ar-tgo), and the length of the corpus (tgo-gn) were significantly smaller in the JIA patients. The average growth direction of the chin was more downward in the JIA patients. A smaller posterior facial height and a higher ratio between the anterior and posterior facial heights were also observed. The differences in craniofacial morphology between the patients and the control group were more pronounced at 35 years of age than at earlier ages. ***Conclusion.*** The typical craniofacial morphological pattern in JIA patients was established between 9 and 12 years of age. From the age of 12 until the age of 35, this morphological pattern remained relatively stable, in contrast to the pattern in the healthy control subjects.

Key Words: *Craniofacial morphology, growth abnormalities, juvenile arthritis*

Introduction

Juvenile idiopathic arthritis (JIA) comprises a heterogenic group of rheumatic diseases. It includes seven subtypes according to the International League of Associations for Rheumatology (ILAR) criteria [1]: systemic arthritis, oligoarthritis (persistent or extended), polyarthritis (rheumatoid factor negative), polyarthritis (rheumatoid factor positive), psoriatic arthritis (PSA), enthesitis-related arthritis (ERA), and undifferentiated arthritis. Oligoarthritis, or pauciarticular arthritis, affects one to four joints, whereas polyarthritis affects five or more joints. The JIA diagnosis is a substitute for the American juvenile rheumatoid arthritis (JRA) diagnosis and the European juvenile chronic arthritis (JCA) diagnosis.

Research has shown that JIA may affect the temporomandibular joint (TMJ) and disturb the growth of the mandible [2–6]. Barriga [7], Bache [8], Kjellberg et al. [9] and Kjellberg [10] have often observed an Angle class II occlusion in combination with a retrognathic mandible and smaller mandibular dimensions (corpus length and ramus height) in JIA patients [2,9,11,12]. In addition, the mandible is often rotated posteriorly and a marked antegonial notching may be apparent, as reported by Rönning et al. [11], Kreiborg et al. [13], and Bjørk & Skieller [14]. The posterior rotation may lead to an anterior open bite [8,10], although Barriga [7] and Jämsä & Rönning [15] found insignificant changes in the overbite during growth. Proclined lower incisors were found by Stabrun [12] and Kjellberg [10].

It is not surprising that most studies on JIA patients are carried out in children and adolescents. Therefore little is known about the long-term development of craniofacial morphology into adult age in

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these patients [3,16]. The objective of the present study was to compare the development of average craniofacial morphology in a group of Norwegian JIA patients with different diagnosis from 6 years to 35 years of age with a matched group of healthy individuals.

Material and methods

Healthy control subjects

The control group with children was taken from "The University of Oslo Craniofacial Growth Archives". Lateral cephalograms, study models, and X-rays were obtained at the ages 6, 9, and 12 years of age. None of the subjects had undergone orthodontic treatment.

The adult controls originated from files of "The University of Oslo Craniofacial Growth Archives" as well. They were enrolled in the archives during their dental education and were assessed for new examinations every 10 years.

All controls, children and adults, were without known or suspected joint disease. They were all selected without regard to skeletal type and malocclusion. They therefore represent a random selection of skeletal types and malocclusions at the various age levels.

JIA patients

The original material comprised 103 Norwegian JIA patients first examined between 1976 and 1979. They included 71 girls and 32 boys examined at 2-year intervals and followed for either 4 or 6 years. The patients were classified according to the American Rheumatoid Association classification criteria for juvenile rheumatoid arthritis, as described by Stabrun et al. [2]. Their onset types were: systemic 13 (13%), pauciarticular 65 (63%), and polyarticular 25 (24%). The mean age of all patients at baseline was 9.0 years (range 2.5–16.4 years) and the average onset age of disease was 5.5 years (range 0.8–14.5 years).

Of the 103 patients, 60 were re-examined 23–31 years after baseline examination and, of these 60 patients, 6 were excluded because of surgery in the craniofacial area. Thus, 54 patients typical of the original material were available for examination in the present study. They included 38 females and 16 males with a mean age at baseline recording of 8.6 years (range 2.8–16.4). The mean age of 33.3 years for the adult controls corresponded well with the JIA group as adults with a mean age of 35.3 years. Age at onset of the disease for the 54 patients in the present study and onset type are given in Table I.

Most of the JIA patients had four or five lateral cephalograms available for analysis. This provided a wide distribution in age at the examinations for each patient. The control group had recordings on average at 6, 9, 12, and 33 years, and the JIA patients were allocated into these age levels. A deviation of ± 1 year at the first three age levels was accepted in the study, whereas all 54 patients were included as adults. At the 6 years age level, 16 patients met the inclusion criteria; at 9 years, 32 patients; and at 12 years, 32 patients. Although an equal number of patients was included at the 9 and 12 years age levels, it is not all the same patients.

The control subjects were matched to each JIA patient in regard to age and gender at the various age levels, and the study design was thus cross-sectional at the different age levels.

The patients were reclassified in accordance with the ILAR classification [1] by rheumatologists at the Department of Rheumatology, Rikshospitalet–Radiumhospitalet Medical Center, Oslo, Norway. Data from one patient were insufficient and could not be reclassified. The remaining 53 patients were classified as: oligoarticular arthritis 12 (23%), extended oligoarticular arthritis 15 (28%), polyarticular arthritis, rheumatoid factor negative 11 (21%), polyarticular arthritis, rheumatoid factor positive 2 (4%), systemic arthritis 6 (11%), enthesitis-related arthritis 6 (11%), and psoriatic arthritis 1 (2%).

At baseline examination, no differences were found between the 54 patients included in the present study and the original 103 patients in regard to onset age, onset type, distribution of gender, number of joints with active arthritis, disease activity, physical function-Health Assessment Questionnaire (HAQ) scores and length of treatment with corticoid steroids.

Orthodontic treatment had been performed in 15 (28%) of the 54 JIA patients. Thirty-nine (72%) of the JIA patients had received no orthodontic treatment.

Most of the lateral cephalograms ($n = 146$) were performed on a Lumex B (Siemens) cephalostat. The distance from the focus to the median plane was 180 cm and the median plane-film distance was 10 cm. A few cephalograms ($n = 3$) were done on a Planmeca Promax Cephalostat, and the images were printed on a Fujifilm DryPix 7000 printer. The subjects were seated in the cephalostat and orientated to the FH plane with the teeth in maximum intercuspidation.

All cephalograms were digitized with an Epson 1680 Pro scanner (Epson, Long Beach, USA) and traced with Facad® software (Ilexis AB, Linkoping; Sweden) by one of the authors. The enlargement factor for each cephalogram was adjusted to 1.0.

Cephalometric landmarks and reference lines are shown in Figures 1 and 2.

Table I. Data on the JIA patients for each age level.

	Age of JIA patients		Onset age of disease		JIA onset type		
Age level	Mean	SD	Mean	SD	1	2	3
6 years ($n=16$)	6.1	0.6	3.6	1.6	$n=2$ (13%)	$n=12$ (75%)	$n=2$ (13%)
9 years ($n=32$)	9.0	0.6	4.7	2.5	$n=4$ (13%)	$n=19$ (59%)	$n=9$ (28%)
12 years ($n=32$)	11.9	0.6	5.6	3.0	$n=1$ (3%)	$n=20$ (63%)	$n=11$ (34%)
35 years ($n=54$)	35.3	3.6	5.6	3.0	$n=6$ (11%)	$n=34$ (63%)	$n=14$ (26%)

JIA onset types: 1 systemic arthritis, 2 pauciarticular arthritis, 3 polyarticular arthritis.

Statistical methods

Prior to tracing the cephalograms, a study of the errors of landmark identification was accomplished and measurements were taken by the principal author (M.F.) in collaboration with an experienced orthodontist (K.B.). Twenty randomly selected cephalograms were analyzed twice by the two observers at 3-week intervals.

The descriptive data included mean and standard deviation for each measurement. The data had a normal distribution and the equality of variance was attended in both groups. Student's *t*-test was used to compare differences between the JIA group and controls at each age level. The level of significance was set at $p \leq 0.05$.

Ethical approval

The study protocol was approved by the Regional Committee for Medical Research Ethics, Southern Norway, and informed consent was obtained from the participants at the follow-up investigation.

Results

The method error study showed an acceptable inter- and intra-rater reliability measured by intra-class correlation (ICC). The inter-rater reliability had an ICC value between 0.95 and 0.99 and the intra-rater reliability an ICC value between 0.88 and 0.99.

Comparison between JIA patients and healthy controls at 6 years of age (n = 16)

At the age of 6 years (Table II), a significantly larger A-n-B angle and more proclined lower incisors were found in the JIA patients than in the healthy controls.

Comparison between JIA patients and healthy controls at 9 years of age (n = 32)

At this age level (Table III), the following measurements showed statistically significant differences between the JIA patients and healthy controls. The

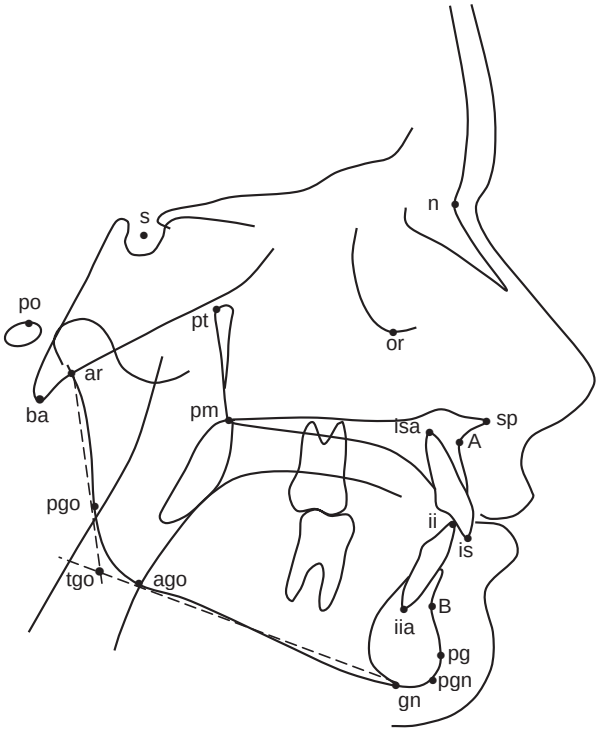

Figure 1. Cephalometric landmarks identified at each cephalogram.

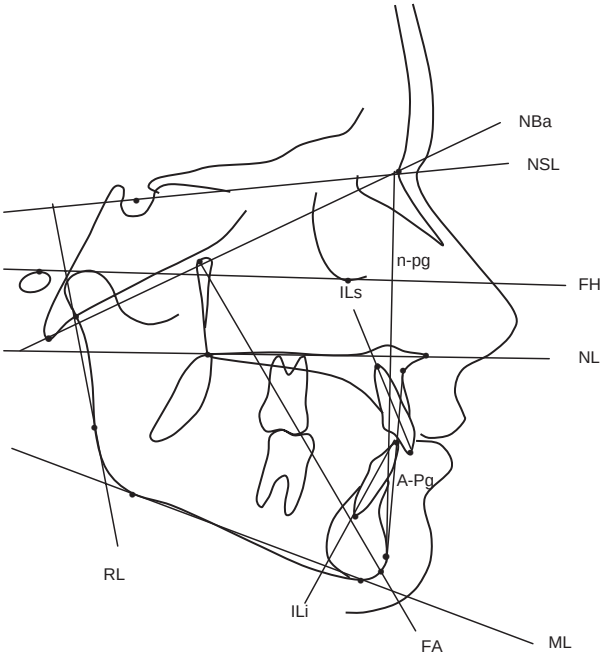

Figure 2. Reference lines identified at each cephalogram.

Table II. Cephalometric measurements for JIA patients and healthy controls at 6 years of age.

		JIA ($n=16$)		Control ($n=16$		
Measurement		Mean	SD	Mean	SD	p-value
Basal sagittal						
n-s-ba	degree	132.1	4.4	133.0	3.3	0.50
s-n-A	degree	81.7	3.4	81.7	3.1	0.89
s-n-B	degree	75.9	3.4	77.8	3.0	0.075
A-n-B	degree	5.9	2.5	4.1	2.2	**0.041**
A ⊥ n-pg	mm	4.4	2.2	3.4	2.0	0.19
NL/NSL	degree	7.9	2.3	7.3	3.4	0.55
ML/NSL	degree	32.9	5.2	32.4	2.8	0.74
ML/NL	degree	25.0	5.4	25.2	3.1	0.88
FA/NBa	degree	92.2	4.1	93.2	3.0	0.36
n-ans ⊥ FH	mm	39.4	2.8	39.2	2.2	0.80
ans-gn ⊥ FH	mm	50.6	3.8	50.9	3.1	0.72
n-ans/ans-gn	%	78.1	5.7	77.2	6.6	0.68
n-gn	mm	91.3	5.3	91.0	3.7	0.82
NSL-tgo	mm	56.5	5.1	57.4	2.6	0.36
n-gn/NSL-tgo	%	162	12	159	7	0.25
Mandibular						
ba-ar	mm	11.1	3.1	10.8	1.6	0.73
ar-gn	mm	82.9	6.4	83.8	2.8	0.60
ar-tgo	mm	35.0	3.8	36.2	1.8	0.22
gn-tgo	mm	57.2	4.6	56.3	2.5	0.39
ar-tgo-gn	degree	126.8	5.4	128.9	4.8	0.17
Dental						
ii ⊥ A-pg	mm	−0.1	2.1	0.1	1.4	0.62
ILi/A-pg	degree	16.5	6.6	13.4	6.2	0.13
ILi/ML	degree	93.5	8.2	88.1	7.2	**0.031**
is ⊥ A-pg	mm	2.9	1.7	2.5	1.8	0.46
ILs/A-pg	degree	21.7	10.6	20.0	8.2	0.47
ILs/NSL	degree	91.8	9.5	92.9	6.5	0.62
ILi/ILs	degree	141.9	15.5	146.6	12.2	0.16

JIA patients had more protrusion and proclination of the maxillary incisors than the control subjects. This was due to a more posterior position of pogonion in the JIA patients than in the controls. The ratio between the anterior and posterior facial heights was higher in the JIA. In addition, the distance ba-ar was greater in the JIA patients.

Comparison between JIA patients and healthy controls at 12 years of age ($n=32$)

At 12 years of age, several measurements were statistically significantly different between the two groups (Table IV) and are described below. A retrognathic position of the mandible was found in the JIA patients, indicated by a smaller s-n-B angle and a higher A-n-B angle. The mandibular dimensions were also smaller in the JIA patients, indicated by the reduced linear measures ar-gn, ar-tgo and gn-tgo. The distance ba-ar was greater in the JIA patients. A relatively steep mandibular plane angle – demonstrated by an increased NSL/ML angle – a more downward than forward orientation of the chin, and an increased mandibular angle were found in the JIA group. The posterior facial height was smaller and the ratio between anterior and posterior facial heights higher in the JIA patients compared to the controls.

The JIA patients had more protruded and proclined upper incisors and more protruded lower incisors.

Comparison between JIA patients and healthy controls at 35 years of age ($n=54$)

Statistically significant differences between the two groups from Table V are described below. The mandible in the JIA patients was in a retrognathic position with a smaller s-n-B angle and a higher A-n-B angle compared to controls. The ML/NSL angle, too, was higher in the JIA group. The indication of the chin orientation was more downward compared to that of the controls.

The length of the mandible (ar-gn) was smaller for the JIA group than for the controls. Also ramus height (ar-tgo) and body length (gn-tgo) were smaller in the JIA patients. At this age level, the distance (ba-ar) was longer in the JIA patients.

The posterior facial height was significantly smaller in the JIA patients than in the controls. Hence, the ratio between the anterior and the posterior facial heights was higher in JIA patients.

Table III. Cephalometric measurements for JIA patients and healthy controls at 9 years of age.

		JIA ($n=32$)		Control ($n=32$)		
Measurement		Mean	SD	Mean	SD	p-value
Basal sagittal						
n-s-ba	degree	131.4	3.9	130.9	3.3	0.58
s-n-A	degree	80.7	3.9	81.0	3.3	0.72
s-n-B	degree	76.3	3.5	77.5	3.0	0.14
A-n-B	degree	4.4	2.5	3.5	2.1	0.13
A ⊥ n-pg	mm	3.5	2.4	2.4	2.2	0.074
NL/NSL	degree	8.4	2.5	7.8	2.3	0.28
ML/NSL	degree	33.9	5.9	31.8	3.3	0.092
ML/NL	degree	25.5	5.6	24.0	3.0	0.18
FA/NBa	degree	90.5	4.2	91.9	3.0	0.17
n-ans ⊥ FH	mm	42.3	2.6	42.8	2.2	0.54
ans-gn ⊥ FH	mm	53.4	3.6	53.4	2.6	0.93
n-ans/ans-gn	%	79.7	5.7	80.3	5.7	0.69
n-gn	mm	97.3	5.0	97.0	3.7	0.84
NSL-tgo	mm	59.4	6.3	61.5	3.2	0.088
n-gn/NSL-tgo	%	165	17	158	8	**0.030**
Mandibular						
ba-ar	mm	12.1	3.8	10.6	2.0	**0.036**
ar-gn	mm	87.3	6.1	88.7	3.8	0.25
ar-tgo	mm	36.1	4.1	37.2	2.2	0.18
gn-tgo	mm	61.4	4.3	61.5	3.5	0.96
ar-tgo-gn	degree	125.1	4.7	126.4	4.4	0.26
Dental						
ii ⊥ A-pg	mm	1.8	1.8	1.3	1.2	0.13
ILi/A-pg	degree	21.3	4.8	21.5	4.0	0.89
ILi/ML	degree	95.6	5.6	94.7	4.9	0.49
is ⊥ A-pg	mm	5.7	1.8	4.9	1.7	**0.031**
ILs/A-pg	degree	31.1	6.3	27.5	5.8	**0.013**
ILs/NSL	degree	103.0	5.4	102.5	5.4	0.73
ILi/ILs	degree	127.5	8.8	131.0	8.2	0.090

Both the upper and lower incisors were protruded and proclined in the JIA patients, and a smaller inter-incisal angle was found compared to healthy controls.

Discussion

The JIA patients in the present study were followed for an average time period of 28 years from childhood, through adolescence and into adult age. To our knowledge, craniofacial morphology in JIA patients has never been followed for such a long period of time.

Major findings

The major findings in the present study were the differences between the JIA patients and the healthy controls that appeared from 9 to 12 years of age. Typically, the JIA patients had a more retrognathic position of the mandible with a relatively steeper mandibular plane angle and a smaller posterior facial height. The mandibular dimensions, such as mandibular length, height of the ramus and length of the corpus, were significantly smaller compared to the control subjects. Most of these differences increased between the two groups from adolescence to 35 years of age, mainly due to further anterior growth rotation of the mandible after the age of 12 years in the control subjects; no anterior growth rotation of the mandible could be observed in the JIA patients. The craniofacial morphology of the JIA patients thus remained fairly stable until the age of 35 years.

Vertical dimensions

The JIA patients had a steeper mandibular plane angle than the healthy controls from 12 years of age. At 35 years of age, the mean differences in the ML/NSL and the ML/NL angle were about 6° and 5° higher, respectively, in JIA patients. The main reason for this difference is thought to be the reduced vertical growth of the mandible when one or both of the mandibular condyles are affected by arthritis. Compensatory mechanisms to camouflage a steep mandibular plane angle include apposition in the gonial region, which results in an antegonial notching [11,13–15] and resorption of the lower surface of the symphysis [14].

A posteriorly rotated mandible in JIA patients may lead to an anterior open bite [8,10]. However, compensatory vertical growth often takes place in the anterior part of the alveolar ridge in the

Table IV. Cephalometric measurements for JIA patients and healthy controls at 12 years of age.

		JIA ($n=32$)		Control ($n=32$)		
Measurement		Mean	SD	Mean	SD	*p*-value
Basal sagittal						
n-s-ba	degree	131.1	4.2	130.3	3.8	0.37
s-n-A	degree	80.8	3.8	82.2	3.4	0.15
s-n-B	degree	76.5	3.4	79.4	3.1	**0.002**
A-n-B	degree	4.3	2.6	2.9	2.3	**0.036**
A ⊥ n-pg	mm	3.7	2.8	1.8	2.6	**0.012**
NL/NSL	degree	8.8	2.7	7.3	2.1	**0.015**
ML/NSL	degree	35.4	5.5	28.7	4.3	**<0.001**
ML/NL	degree	26.6	5.9	21.4	4.1	**<0.001**
FA/NBa	degree	89.2	3.4	93.0	3.5	**<0.001**
n-ans ⊥ FH	mm	45.6	2.9	45.8	2.3	0.77
ans-gn ⊥ FH	mm	57.2	4.9	56.5	3.6	0.51
n-ans/ans-gn	%	80.1	6.0	81.4	6.9	0.41
n-gn	mm	104.2	6.7	102.8	4.1	0.28
NSL-tgo	mm	63.2	6.6	68.0	4.2	**0.002**
n-gn/NSL-tgo	%	166	14	152	10	**<0.001**
Mandibular						
ba-ar	mm	12.9	3.4	10.1	1.8	**0.001**
ar-gn	mm	92.3	6.6	95.5	4.4	**0.033**
ar-tgo	mm	38.5	4.5	41.4	3.3	**0.009**
gn-tgo	mm	64.5	5.2	67.4	3.7	**0.007**
ar-tgo-gn	degree	126.0	6.8	121.2	5.4	**0.001**
Dental						
ii ⊥ A-pg	mm	2.4	1.7	1.5	1.7	**0.016**
ILi/A-pg	degree	23.4	4.8	23.2	3.8	0.83
ILi/ML	degree	95.8	5.1	96.4	5.0	0.58
is ⊥ A-pg	mm	6.1	2.1	4.6	2.1	**0.003**
ILs/A-pg	degree	28.2	6.3	24.3	6.6	**0.020**
ILs/NSL	degree	100.5	6.9	102.5	5.9	0.24
ILi/ILs	degree	128.4	8.9	132.4	8.8	0.053

mandible, and the incisors of the mandible and the maxilla might over-erupt to camouflage this tendency. No difference in the lower anterior facial height was found between JIA patients and controls in this study, at any age levels – this in contrast to findings by Ronning et al. [11] and by Sidiropoulou-Chatzigianni et al. [17].

Growth directions

The growth direction of the chin (FA/NBa) in JIA patients and controls during craniofacial maturation was different from 12 years of age. The direction of the facial axis in JIA patients was more downward than forward, i.e. a consequence of reduced horizontal and vertical mandibular growth in JIA patients. As the vertical component of the condylar growth is reduced, a posterior rotation of the mandible takes place, with the centre of rotation at the molars [18]. With a reduced horizontal component, no anterior growth rotation of the mandible takes place.

The distance from basion to articulare is a measurement of the position of the mandibular condyle in relation to the cranial base. In the JIA patients, this distance was on average greater than in controls, as also observed by Stabrun [12]. A possible explanation is that the smaller mandible is brought into a more anterior position to reduce the retrognathism.

Mandibular dimensions

From the age of 12 years, mandibular dimensions such as total length (ar-gn), height of ramus (ar-tgo), and corpus length (tgo-gn) were significantly smaller in the JIA patients compared to the control subjects. The reduced mandibular dimensions are probably a combined effect of impaired condylar growth due to arthritis and adverse effects of corticosteroid medication [2,8,19]. Larheim & Haanaes [3] defined micrognathia as the mean distance (ar-gn) –2 SD in the present study: $\leq$92.9 mm. In adult patients in the present study, 18% of the JIA patients could be classified with micrognathia compared to 2% in the control group.

Dental occlusion and sagittal relationship

In the present study, 56% of the adult JIA patients were classified with a distal basal jaw relationship (A-n-B angle >4 degrees) compared to 26% in the control group. This is slightly lower than observed by Twilt et al. [20] (67%). No information was given of

Table V. Cephalometric measurements for JIA patients and healthy controls at 35 years of age.

		JIA ($n=54$)		Control ($n=54$)		
Measurement		Mean	SD	Mean	SD	*p*-value
Basal sagittal						
n-s-ba	degree	131.6	4.7	128.2	3.7	**<0.001**
s-n-A	degree	81.2	3.9	82.6	3.3	0.079
s-n-B	degree	76.6	3.8	79.9	3.5	**<0.001**
A-n-B	degree	4.7	2.6	2.6	2.3	**<0.001**
A ⊥ n-pg	mm	4.0	3.2	0.7	2.6	**<0.001**
NL/NSL	degree	9.2	3.5	7.7	3.0	**0.028**
ML/NSL	degree	33.8	7.0	27.7	5.3	**<0.001**
ML/NL	degree	24.7	6.1	19.9	5.6	**<0.001**
FA/NBa	degree	88.7	4.6	92.6	3.8	**<0.001**
n-ans ⊥ FH	mm	49.5	3.6	49.8	3.1	0.59
ans-gn ⊥ FH	mm	63.8	6.6	63.3	5.2	0.67
n-ans/ans-gn	%	78.2	7.5	79.3	7.9	0.44
n-gn	mm	114.6	9.2	113.5	6.2	0.46
NSL-tgo	mm	71.4	9.1	77.5	6.8	**<0.001**
n-gn/NSL-tgo	%	162	17	147	12	**<0.001**
Mandibular						
ba-ar	mm	12.4	3.9	9.1	2.9	**<0.001**
ar-gn	mm	100.9	8.3	106.1	6.6	**0.001**
ar-tgo	mm	45.0	6.0	47.8	5.4	**0.010**
gn-tgo	mm	70.8	5.7	73.6	5.1	**0.014**
ar-tgo-gn	degree	120.0	6.1	120.6	5.4	0.60
Dental						
ii ⊥ A-pg	mm	2.7	2.5	0.9	2.4	**<0.001**
ILi/A-pg	degree	25.0	4.7	23.5	5.1	0.14
ILi/ML	degree	98.3	6.6	94.9	6.3	**0.018**
is ⊥ A-pg	mm	6.4	2.7	4.0	2.4	**<0.001**
ILs/A-pg	degree	25.3	7.1	19.8	5.9	**<0.001**
ILs/NSL	degree	98.2	6.4	100.7	6.3	**0.042**
ILi/ILs	degree	129.7	10.4	136.7	10.0	**0.001**

the age of the JIA subjects in that study. A typical finding reported by others is an increased number of Angle class II bite in JIA patients because of the smaller mandibular dimensions [9,12].

Dental compensation in the form of proclined lower incisors has been reported by Stabrun [12] and by Kjellberg [10]. In the present study, this was generally pronounced at the age of 35 years. As adults, no statistically significant differences were found in analysis of the cephalograms between the JIA patients with or without orthodontic treatment.

Rheumatological factors

The average age of JIA patients at disease onset varied between the groups (Table I). Disease duration has been demonstrated as being an important factor in the severity of growth disturbances [2,9] and may partly explain the larger average differences in craniofacial morphology between the 12 and 6-year-old patients.

Onset type has also been reported to affect craniofacial development. Polyarticular onset was reported by Mericle et al. [21] to lead to more severe craniofacial disturbances than the pauciarticular onset type. This may have influenced the results toward a more "normal" craniofacial morphology at 6 years of age.

The present study comprised JIA patients in their growth period during the 1970s and 1980s. Medication of JIA patients has changed markedly in the past 30 years. Today, individual medication regimens are used according to the subtype of JIA present [22]. Perhaps many of the adverse effects of medication on growth demonstrated in the present study have been reduced with current medication.

The study groups

In the present study, the average craniofacial morphology of a mixed group of JIA patients at the ages of 6, 9, 12, and 35 years was studied. This was done rather than study average changes in the different subtypes of JIA according to disease onset because of the low number of individuals in the many subgroups. This may limit the information obtained in the study, but will be further addressed in a subsequent communication. Most important in studies like this is a reliable control group of healthy individuals. This was available from the Oslo craniofacial growth archives for the ages studied. Furthermore, the control groups were composed of

the same relative distribution of gender as in the JIA group.

Conclusions

The typical craniofacial morphological pattern in this mixed group of JIA patients was established between 9 and 12 years of age. From age 12 until age 35, this morphological pattern remained relatively stable, in contrast to that of the healthy control subjects, where the mandible rotated more anteriorly with age.

The differences in craniofacial morphology between the patients and the control group were more pronounced at 35 years of age than at earlier ages. In adults, the main differences of clinical interest were a steeper mandibular plane, a smaller and more retrognathic mandible, and more proclined mandibular incisors in the JIA patients than in the healthy controls.

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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