Long-Term Follow-Up of Orthognathic Surgery in 19 Patients with Juvenile Idiopathic Arthritis

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ABSTRACT

Objectives: Dentofacial deformity following juvenile idiopathic arthritis with temporomandibular joint involvement is associated with functional, aesthetic, and psychosocial impairment. Surgical treatment may involve combinations of orthognathic surgery. The aims of this retrospective study were to assess orofacial symptoms, functional and aesthetic status, and stability after orthognathic surgery.
Material and Methods: Nineteen patients with juvenile idiopathic arthritis of the temporomandibular joint (TMJ) and dentofacial deformities were included. All patients were treated with combinations of bilateral sagittal split osteotomy, Le Fort I and/or genioplasty, between September 10, 2007 and October 17, 2017. Analysis of patient symptoms and clinical registrations, and frontal/lateral cephalograms was performed pre- and postoperative and long-term (mean: 3.8 and 2.6 years, respectively).
Results: Patients experienced no changes in orofacial symptoms or TMJ function, and stable normalisation of horizontal and vertical incisal relations at long-term (horizontal overbite; vertical overbite: $P < 0.05$). Mandibular lengthening was achieved postoperatively (from mean 79.7 to 87.2 mm; $P = 0.004$) and was stable. Sella-nasion to A point (SNA) and sella-nasion to B point (SNB) angles increased postoperatively (SNA, mean 79.9° to 82.8°; $P = 0.022$ and SNB, mean 73.9° to 77.8°; $P = 0.003$), however, largely reverted to preoperative status at long-term.
Conclusions: Orthognathic surgery normalized incisal relations while providing stable mandibular lengthening without long-term deterioration of temporomandibular joint function or orofacial symptoms. No long-term effect on jaw advancements was observed.

Keywords: cephalometry; dentofacial deformities; juvenile arthritis; orthognathic surgery; temporomandibular joint disorders.

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INTRODUCTION

Juvenile idiopathic arthritis (JIA) is the broad term for a group of subtypes of chronic arthritis in children. The reported incidence of JIA in Scandinavia is 15 per 100,000 children/year [1] and the temporomandibular joint (TMJ) is reportedly frequently involved: magnetic resonance imaging (MRI) studies report 43 to 87% incidence rate of TMJ involvement among patients with JIA [2-4].

TMJ JIA is associated with orofacial pain and radiologic TMJ deformation [5] as well as dentofacial deformity [6-8] with negative psychosocial and functional issues [5,9-13]. Severity is dependent on multiple factors, e.g. age at onset, subtype of disease, and whether TMJ involvement is uni- or bilateral.

Treatment is individualised and includes combinations of medical systemic treatment [14] with supportive/interceptive orthopaedic devices during growth [15-17] and possible intra-articular corticosteroid TMJ injection [18]. Patients with severe dentofacial deformities may require orthognathic surgical treatment (OS), e.g. bilateral sagittal split osteotomy (BSSO), Le Fort I and genioplasty, which is generally not performed until skeletal maturation is reached and disease is quiescent. In select cases, patients may require mandibular distraction osteogenesis (MDO) alone or as an adjunct to other OS, e.g. in severe unilateral cases, in large bilateral advancement cases, and for early intervention.

Few studies are available on the outcomes of OS in patients with JIA, and suffer from small sample sizes with no long-term follow-up [19]. In particular, very few studies have examined the effect of OS on symptoms and TMJ function.

The aims of this retrospective study were to analyse short- and long-term: 1) orofacial symptoms and temporomandibular joint function and 2) dentofacial morphology and stability following bilateral sagittal split osteotomy, Le Fort I and/or genioplasty to correct dentofacial deformities in patients with juvenile idiopathic arthritis of the temporomandibular joint.

MATERIAL AND METHODS

Study design and patients

In this retrospective two-center study, 19 patients with JIA and TMJ involvement were treated between September 10, 2007 and October 17, 2017 with one- or two-jaw OS for dentofacial deformities (Table 1 and Figure 1). All patients had pre- and postsurgical orthodontics.

Table 1. Patient, examination and treatment variables

		N	Mean (SD)	Range
Age at surgery				
Years		19	22.2 (5.6)	16.1 - 32.6
Gender				
Male		2 (11%)	-	-
Female		17 (89%)	-	-
TMJ involvement				
Unilateral		4 (21%)	-	-
Bilateral		15 (79%)	-	-
Antirheumatic medication				
No medication		6	-	-
NSAID mono		4	-	-
MTX mono		5	-	-
NSAID + TMJ steroid		1	-	-
Biologics + MTX		1	-	-
Biologics + MTX + NSAID		1	-	-
Biologics + MTX + NSAID + TMJ steroid		1	-	-
Total		19	-	-
Timing of examinations (months)				
T1	Anamnestic and clinical	17	31.9 (18.3)	6.3 - 69.7
	Radiologic	16	5 (5.6)	0.2 - 15.8
T2	Anamnestic and clinical	14	10.3 (5.7)	1 - 20
	Radiologic	16	0.9 (1.7)	0.2 - 6.2
T3	Anamnestic and clinical	16	45.8 (22.6)	14.1 - 93
	Radiologic	11	31.2 (20.4)	9.6 - 68
Surgical treatment				
Genioplasty		2	-	-
Le Fort I		3	-	-
Le Fort I + genioplasty		1	-	-
Le Fort I + BSSO		6	-	-
Le Fort I + BSSO + genioplasty		4	-	-
BSSO		1	-	-
BSSO + genioplasty		2	-	-
Total		19	-	-

N = number of patients; SD = standard deviation; NSAID = nonsteroidal anti-inflammatory drugs; MTX = methotrexate; TMJ = temporomandibular joint; TMJ steroid = intraarticular steroid injection in TMJ; T1 = preoperative examination; T2 = postoperative examination; T3 = long-term examination; BSSO = bilateral sagittal split osteotomy.

The patients were treated at the Department of Oral and Maxillofacial Surgery, Aarhus University Hospital, Denmark (14 patients) and at the Department of ENT & Division of Oral and Maxillofacial Surgery, University Hospital North-Norway, Tromsø, Norway (5 patients).

Ethical approval was given by the respective heads of departments, since no intervention testing was performed, at Aarhus University Hospital and

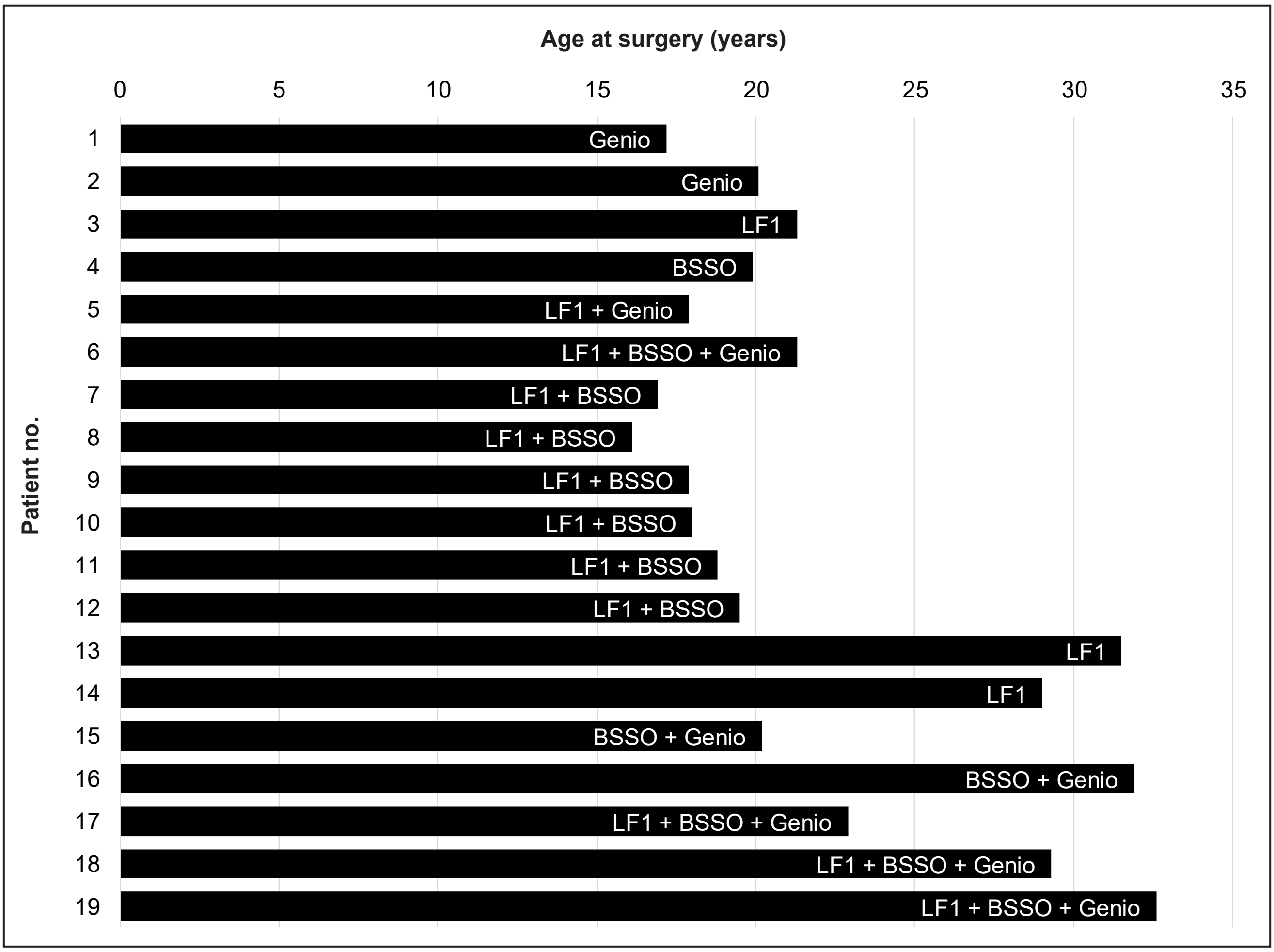

Figure 1. Individual treatment data.
LF1 = Le Fort I; BSSO = bilateral sagittal split osteotomy; Genio = genioplasty.

University Hospital of North Norway. Approval by the Norwegian Data Protection Authority was granted, reference number 2014/1103.

Patient informed consents were signed and obtained as necessary.

Inclusion criteria were: 1) JIA diagnosis according to the International League of Associations for Rheumatology (ILAR) criteria [20]; 2) TMJ involvement (uni- or bilateral); 3) clinical remission; 4) clinical and radiological dentofacial deformity (mandibular retrognathia and/or facial asymmetry) [21]; and 5) subjective and objective acceptable-functioning TMJs.

Disease remission was evaluated by a specialist in paediatric rheumatology according to the provisional criteria of the American College of Rheumatology (ACR) [22]. Clinical and radiologic TMJ stability was evaluated by a trained specialist in orthodontics in Denmark (T.K.P.) or by a specialist in oral and maxillofacial surgery (OMFS) in Norway (P.F.).

Use of anti-rheumatic medication did not constitute an exclusion criterion (Table 1). However, in Denmark methotrexate and biologics were discontinued 1 week prior to surgery and 2 weeks postoperatively, while, in Norway only biologics were discontinued. No patients had prior surgical treatment of the TMJ, whereas 2 were previously treated with intra-articular orticosteroid injections.

Examinations

Clinical and radiologic examinations were performed by trained specialists (P.F., T.K.P., S.E.N.) preoperatively (T1), shortly after primary OS (T2) and at long-term follow-up (T3). The focus of examinations was orofacial symptoms, clinical findings and cephalometric analysis, and examinations complied with TMJ recommendations [23].

Details of the timing of examinations and radiographs are provided in Table 1.

Orofacial symptoms included headache, restricted mouth opening, affected chewing ability and TMJ pain/noises.

Functional clinical registrations included TMJ pain on palpation, crepitation, deviation on opening, maximum interincisal opening capacity (MIO), laterotrusion, protrusion, and pain on palpation of masseteric and temporal muscles.

Clinical morphometric registrations were facial asymmetry, chin deviation, horizontal overbite (HOB) and vertical overbite (VOB).

Radiologic examinations included frontal and lateral cephalograms; conventional in natural head position and calibrated using a fixed length ruler or chain, or constructed from cone-beam computed tomography's (CBCTs) with orientation according to the Frankfurt horizontal plane.

Cephalometric analysis

Anatomic landmarks, reference lines and morphometric measurements are depicted in Figure 2 and 3 with definitions of landmarks in Table 2. Frontal and lateral cephalograms were digitised, calibrated and analysed using Dolphin Imaging software version 11.95.8.64 (Dolphin Imaging & Management Solutions; Los Angeles, California, USA). Cephalometric analysis was performed by an OMFS specialist (P.F.) and resident in OMFS (H.K.).

Posterior facial symmetry was expressed by ramus height ratio as the ratio between the most and the least severely affected facial height in frontal cephalograms.

Surgery

Surgical procedures comprising BSSO, Le Fort I and genioplasty were planned and performed according to conventional surgical principles and techniques with rigid fixation as described by Proffit et al. [24]. A final fixed maxillary splint and guiding elastics were used for BSSO, Le Fort I and bimaxillary procedures. Bimaxillary procedures were performed mandible-first using an intermediary splint.

Statistical analysis

Data was handled and analysed using IBM SPSS® (Statistical Package of Social Sciences Statistics) version 20 software (SPSS, Inc.; Chicago, Illinois, USA). Descriptive statistics (mean and standard deviation [SD], range and frequency, percentage) were used to summarise and analyse changes in orofacial symptoms, clinical and cephalometric characteristics.

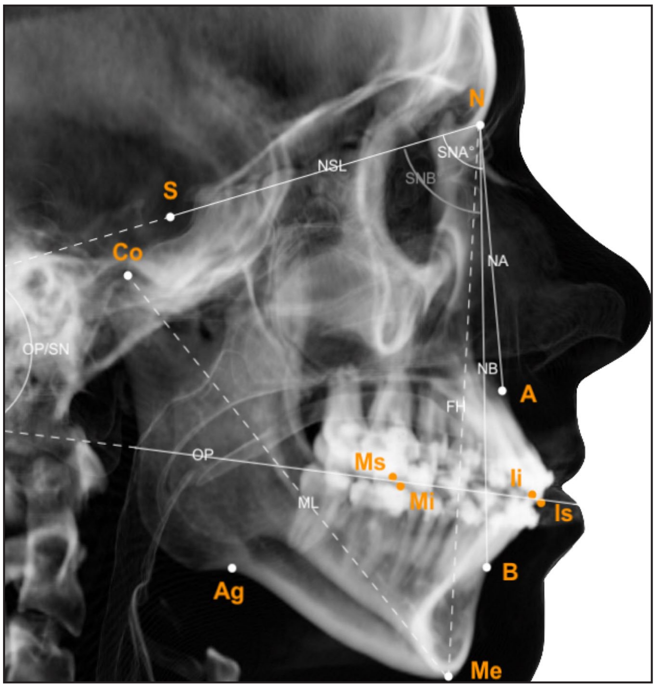

Figure 2. Lateral cephalogram.
Landmarks: S = sella; N = nasion; A = A-point; B = B-point; Is = incision superior; Ms = molar superior; Ii = incision inferior; Mi = molar inferior; Me = menton; Co = condylion; Ag = antegonion.
Reference lines: NSL = nasion-sella line; NA = nasion-A-point line; NB = nasion-B-point line; OP = occlusal plane.
Measures - linear: FH = anterior facial height; ML = mandibular length.
Measures - angular: SNA = sella-nasion-A-point; SNB = sella-nasion-B-point; OP/SN = occlusal plane/SN angle.

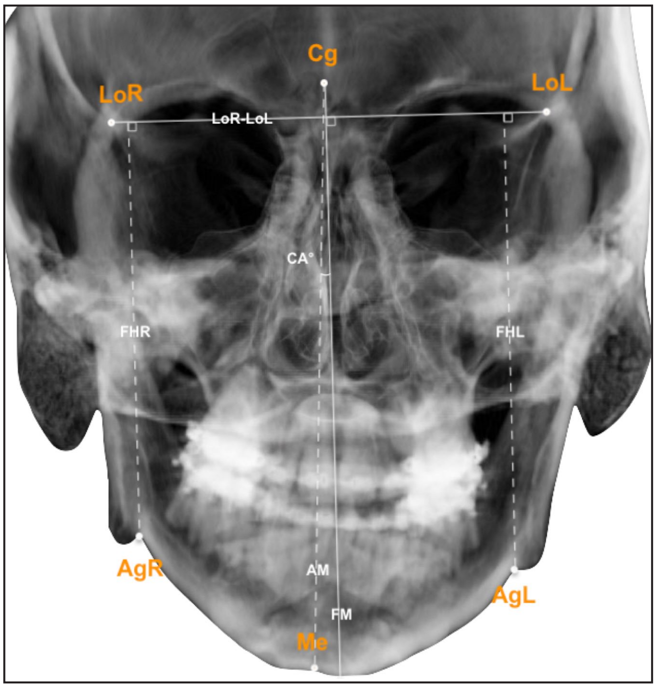

Figure 3. Frontal cephalogram.
Landmarks: Cg = crista galli; LoR = lateroorbitale point, right; LoL = lateroorbitale point, left; Me = menton; AgR = antegonion, right; AgL = antegonion, left.
Reference lines: LoR-LoL = lateroorbital line; FM = facial midline; AM = actual midline.
Measures - linear: FHR = facial height, right = distance from AgR perpendicular to LoR-LoL; FHL = facial height, left = distance from AgL perpendicular to LoR-LoL.
Measures - angular: CA = chin asymmetry = deviation of AM from FM.

Table 2. Definitions of cephalometric landmarks

	Definition	Abbreviation
Lateral landmarks		
Sella	Centre of sella turcica	S
Nasion	Most anterior point of nasofrontal suture	N
A-point	Deepest point on curvature of anterior maxilla between ANS and alveolar process	A
B-point	Deepest point on curvature of anterior mandible between Infradentale and Pogonion	B
Incision superior	Midpoint of incisal edge, most prominent upper central incisor	Is
Molar superior	Mesiofacial cusp of upper first molar	Ms
Incision inferior	Midpoint of incisal edge, most prominent lower central incisor	Ii
Molar inferior	Mesiofacial cusp of lower first molar	Mi
Menton	Most inferior point of mandibular symphysis in midsagittal plane	Me
Condylion	Most superior point of condyle	Co
Antegonion	Highest point on antegonial notch	Ag
Lateral reference lines		
Nasion-sella line	Line through N and S	NSL
Nasion-A-point line	Line through N and A	NA
Nasion-B-point line	Line through N and B	NB
Occlusal plane	Line bisecting cusp height of first permanent molars and bisecting incisal overbite	OP
Frontal landmarks		
Crista galli	Crista galli of the ethmoid	Cg
Lateroorbitale point, right	Intersecting point between lateral orbital contour and oblique orbital line, right side	LoR
Lateroorbitale point, left	Intersecting point between lateral orbital contour and oblique orbital line, left side	LoL
Menton	Most inferior point of mandibular symphysis in midsagittal plane	Me
Antegonion, right	Highest point of antegonial notch, right side	AgR
Antegonion, left	Highest point of antegonial notch, left side	AgL
Frontal reference lines		
Lateroorbital line	Line through LoR and LoL	LoR-LoL
Facial midline	Line perpendicular to LoR-LoL through Cg	FM
Actual midline	Line through Cg and Me	AM

Distributions of continuous variables were assessed for normality using the Shapiro-Wilk test. Fischer's exact test and Chi-square test were used to detect significance in differences in dichotomous registrations. Differences in continuous numeric variables were tested for significance using paired-samples t-test (normally distributed data) or Wilcoxon signed-ranks test (not normally distributed data). P-values less than 0.05 were considered significant.

Data concerning anterior facial height, HOB, VOB, ramus height ratio and chin asymmetry were not included at T2 because patients wore occlusal splints.

RESULTS

The mean age at surgery was 22.2 years. Details on patients and surgical treatments are provided in Table 1 and Figure 1. Datasets were in a few cases incomplete, see N in Tables 1, 3 and 4.

Reported symptoms

Details on reported symptoms are shown in Table 3 and Figure 4. No significant changes of orofacial symptoms were seen from T1-T2 and T2-T3.

Clinical findings

Short-term impairment of TMJ function was observed postoperatively from T1-T2 (MIO, mean 42.9 to 38.5 mm; laterotrusion, mean 7.8 to 6.9 mm; protrusion, mean 6.5 to 5.8 mm), however, changes were not statistically significant. Function returned to near-preoperative status following significant improvements in MIO and laterotrusion between T2-T3 ($P_{T2/T3}$ = 0.029 and 0.039, respectively), and no significant long-term effect was observed on TMJ function (T1-T3). The detailed results are shown in Table 3, Figure 5 and 6.

Table 3. Orofacial symptoms and clinical findings

	T1 (n = 17)			$P_{T1/T2}$	T2 (n = 14)			$P_{T2/T3}$	T3 (n = 16)			$P_{T1/T3}$
	N (%)	Mean (SD)	Range		N (%)	Mean (SD)	Range		N (%)	Mean (SD)	Range	
Reported orofacial symptoms												
Headache	5 (29)	-	-	0.242[b]	7 (47)	-	-	0.156[b]	4 (25)	-	-	1.000[a]
Opening restricted	4 (24)	-	-	1.000[a]	3 (20)	-	-	1.000[a]	3 (19)	-	-	1.000[a]
Chewing restricted	7 (41)	-	-	0.28[a]	3 (20)	-	-	1.000[a]	4 (25)	-	-	0.325[b]
Chewing painful	6 (35)	-	-	1.000[a]	4 (27)	-	-	0.378[a]	2 (13)	-	-	0.225[a]
TMJ clicking	3 (18)	-	-	1.000[a]	2 (13)	-	-	0.657[a]	4 (25)	-	-	0.688[a]
Clinical findings - morphology												
Facial asymmetry	13 (76)	-	-	0.441[a]	8 (53)	-	-	0.464[b]	7 (41)	-	-	0.055[b]
Chin deviation	11 (65)	-	-	0.224[b]	6 (40)	-	-	0.51[b]	5 (29)	-	-	0.055[b]
Horizontal overbite, mm	-	4.4 (3.3)	1 - 13	0.016[d]*	-	2.3 (1)	1 - 5	0.618[d]	-	2.5 (1)	1 - 5	0.004[c]*
Vertical overbite, mm	-	-0.6 (2.7)	-7 - 3	0.004[c]*	-	1.6 (1.5)	-2 - 3	0.088[d]	-	1.3 (1.5)	-2 - 4	0.027[d]*
Clinical findings - function												
TMJ - Pain at palpation - Unilateral	2 (11)	-	-	1.000[a]	1 (7)	-	-	1.000[a]	1 (6)	-	-	1.000[a]
TMJ - Pain at palpation - Bilateral	4 (22)	-	-	0.355[a]	1 (7)	-	-	1.000[a]	2 (12)	-	-	0.656[a]
TMJ - Crepitation - Unilateral	2 (11)	-	-	0.488[a]	0	-	-	1.000[a]	0	-	-	0.485[a]
TMJ - Crepitation - Bilateral	2 (11)	-	-	1.000[a]	2 (13)	-	-	1.000[a]	2 (12)	-	-	1.000[a]
TMJ - Opening deviation	8 (44)	-	-	0.293[b]	4 (27)	-	-	0.709[a]	6 (35)	-	-	0.579[b]
TMJ - MIO, mm	-	42.9 (5.7)	30 - 53	0.062[d]	-	38.5 (7.6)	20 - 53	0.029[c]*	-	40.3 (4.8)	28 - 48	0.333[d]
TMJ - Laterotrusion, mm	-	7.8 (3.1)	0 - 15	0.089[d]	-	6.9 (1.7)	3 - 10	0.039[c]*	-	7.6 (2)	3 - 12	0.808[d]
TMJ - Protrusion, mm	-	6.5 (2.7)	2 - 12	0.130[c]	-	5.8 (2.1)	2 - 9	0.147[b]	-	5.9 (1.7)	3 - 8.5	0.524[c]
Muscles - Pain at palpation - Masseter - Unilateral	3 (17)	-	-	0.607[a]	1 (7)	-	-	1.000[a]	2 (12)	-	-	1.000[a]
Muscles - Pain at palpation - Masseter - Bilateral	5 (28)	-	-	0.698[a]	3 (20)	-	-	0.642[a]	2 (12)	-	-	0.398[a]
Muscles - Pain at palpation - Temporalis - Unilateral	1 (6)	-	-	1.000[a]	0	-	-	1.000[a]	1 (6)	-	-	1.000[a]
Muscles - Pain at palpation - Temporalis - Bilateral	4 (22)	-	-	0.344[a]	1 (7)	-	-	1.000[a]	1 (6)	-	-	0.335[a]

[a]Fischer's exact test for intragroup differences; [b]Chi-square test for intragroup differences; [c]Paired-samples t-test for intragroup differences; [d]Wilcoxon signed-ranks test for intragroup differences.

*Statistically significant (P < 0.05).

T1 = preoperative examination; T2 = postoperative examination; T3 = long-term examination; N = number of patients; SD = standard deviation; TMJ = temporomandibular joint; MIO = maximum interincisal opening capacity.

Table 4. Cephalometric analysis

	T1		$P_{T1/T2}{}^{a}$	T2		$P_{T2/T3}{}^{a}$	T3		$P_{T1/T3}{}^{a}$
	Mean (SD)	Range		Mean (SD)	Range		Mean (SD)	Range	
Lateral headfilms	N = 15			N = 15			N = 11		
SNA angle, (°)	79.9 (4.6)	72.4 - 89.8	0.022*	82.8 (4.5)	74 - 90	0.054	79.5 (4.3)	73.3 - 89.7	0.707
SNB angle, (°)	73.9 (5.2)	66.4 - 85.3	0.003*	77.8 (4.3)	70.9 - 86	0.202	75.5 (3.9)	70.1 - 83.1	0.124
Occlusal plane/SN angle, (°)	24.5 (6.5)	9 - 32	0.079	20.7 (5.9)	10.3 - 30	0.342	21 (4.2)	13.7 - 26.3	0.229
Anterior facial height, mm	110.6 (6.4)	100.4 - 124.6	-	-	-	-	109.3 (6.1)	99.9 - 117.9	0.904
Mandibular length, mm	79.7 (12.2)	62.4 - 103.6	0.004*	87.2 (10.9)	75.3 - 113	0.129	89.8 (15.3)	72.4 - 113.1	0.056
Horizontal overbite, mm	5.3 (2.3)	1.8 - 9.2	-	-	-	-	2.7 (1.2)	1.3 - 5.4	0.004*
Vertical overbite, mm	-0.7 (3.1)	-8 - 2.9	-	-	-	-	1.5 (1.8)	-2 - 3.6	0.219
Frontal headfilms	N = 10		-	-		-	N = 8		-
Ramus height, ratio	-	0.96	-	-	-	-	0.96 (0.05)	0.88 - 1	0.221
Chin asymmetry, (°)	-	1.3	-	-	-	-	1.1 (0.4)	0.5 - 1.7	0.32

[a]Paired-samples t-test for intragroup differences.

*Statistically significant (P < 0.05).

T1 = preoperative examination; T2 = postoperative examination; T3 = long-term examination; N = number of patients; SD = standard deviation; SN = sella-nasion; SNA = sella-nasion to A point; SNB = sella-nasion to B point.

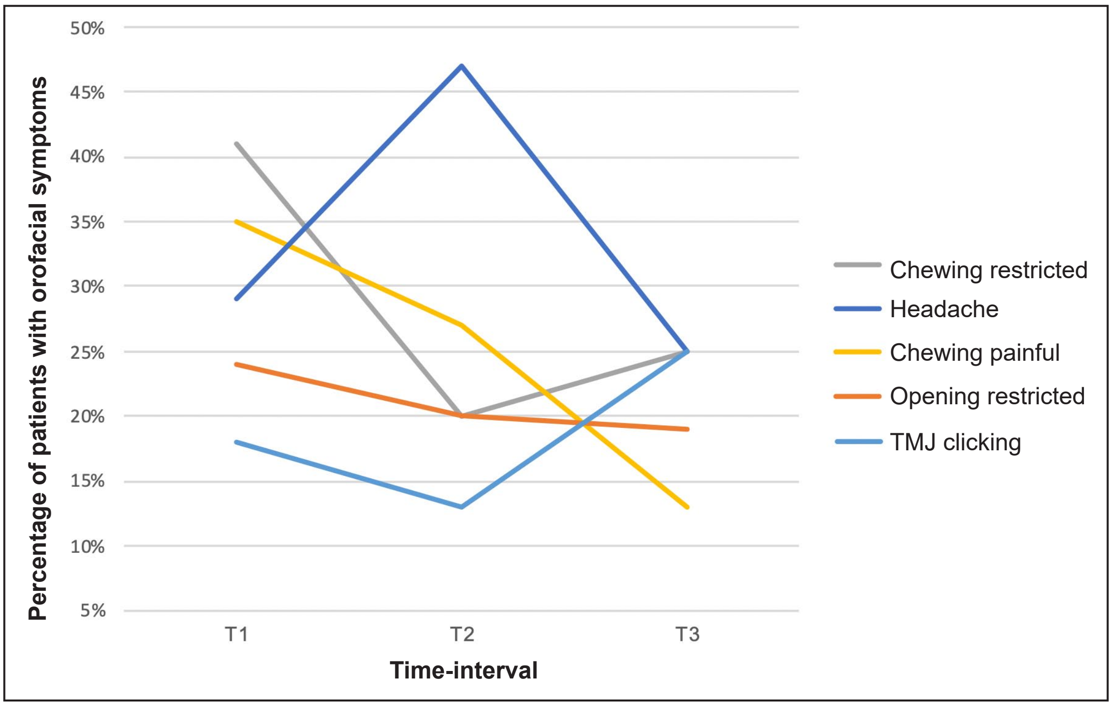

Figure 4. Orofacial symptoms.
T1 = preoperative examination; T2 = postoperative examination; T3 = long-term examination; TMJ = temporomandibular joint. Changes were not statistically significant.

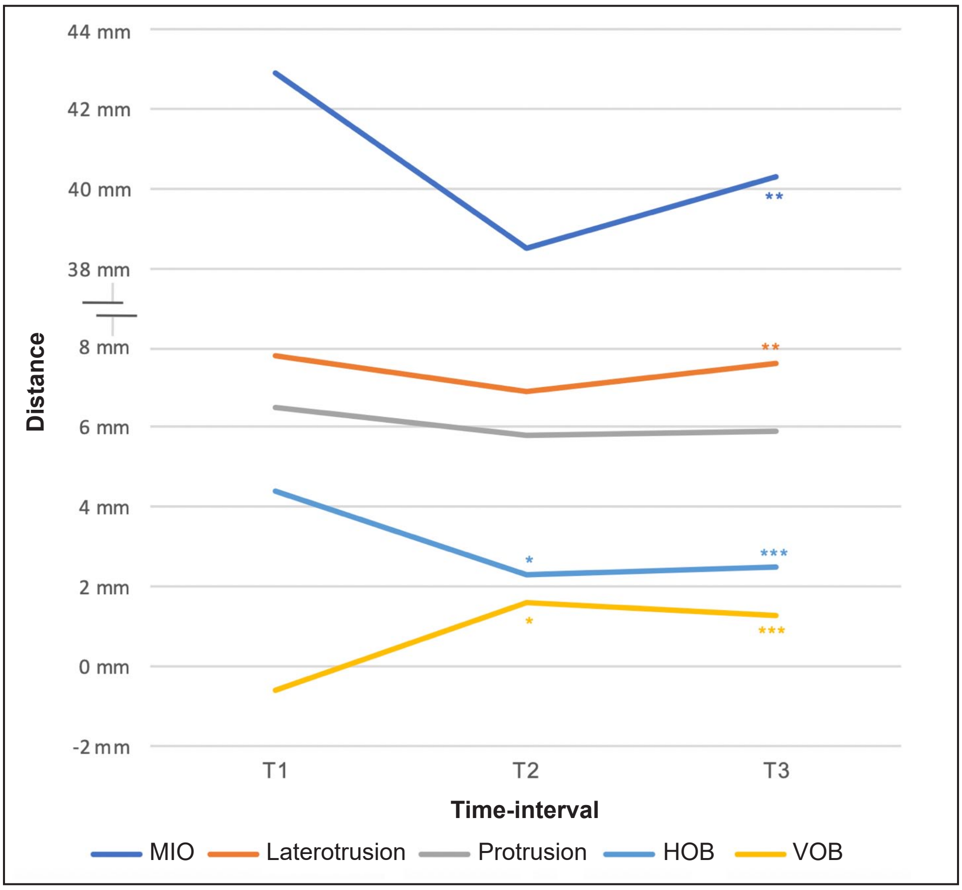

Figure 5. Temporomandibular joint function, horizontal overbite (HOB) and vertical overbite (VOB).
T1 = preoperative examination; T2 = postoperative examination; T3 = long-term examination; MIO = maximum interincisal opening.
*$P_{T1/T2} < 0.05$, Wilcoxon signed-ranks test for differences in HOB within groups, paired-samples t-test for differences in VOB within groups.
**$P_{T2/T3} < 0.05$, paired-samples t-test for differences in MIO and laterotrusion within groups.
***$P_{T1/T3} < 0.05$, paired-samples t-test for differences in HOB within groups, Wilcoxon signed-ranks test for differences in VOB within groups.

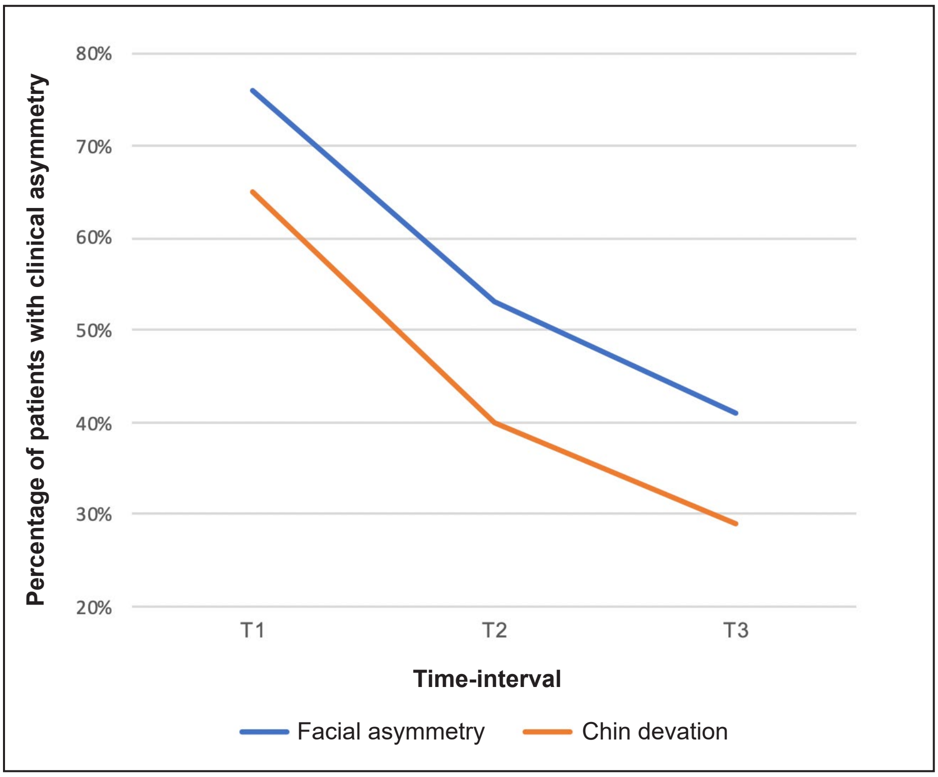

Figure 6. Clinical assessed asymmetry.
T1 = preoperative examination; T2 = postoperative examination; T3 = long-term examination.
Improvements were not statistically significant.

TMJ or muscular pain on palpation was unchanged.

Proportions of patients with facial asymmetry and chin deviation did not change significantly, however, short- and long-term reductions of both were observed. Incidences decreased between T1-T3 ($P_{T1/T3}$ = 0.055 in each case) from 76% and 65%, respectively, to 41% and 29%.

Preoperative class II incisal relations (HOB, mean 4.4 mm) and open bite (VOB, mean -0.6 mm) were normalised postoperatively (HOB, mean 2.3 mm; $P_{T1/T2}$ = 0.016 and VOB, mean 1.6 mm; $P_{T1/T2}$ = 0.004). Changes were stable and significant at long-term ($P_{T1/T3}$ = 0.004 and 0.027, respectively).

Cephalometric analysis

Sella-nasion to A point (SNA) point angle was preoperatively on average 79.9° (range 72.4 to 89.8°) at T1, and significant postoperative advancements were achieved (SNA angle, mean 82.8°; range from 74 to 90°; $P_{T1/T2}$ = 0.022) at T2. Results indicate long-term return to preoperative status at T3 and no significant long-term sagittal effect. Le Fort I procedures were performed in 14 of 19 patients. Results are shown in detail in Table 4, Figure 7 and 8.

The preoperative sella-nasion to B point (SNB) angle at T1 was mean 73.9° (range 66.4 to 85.3°), and significant postoperative advancements were observed at T2 (SNB angle, mean 77.8°; range 70.9 to 86°; $P_{T1/T2}$ = 0.003). Long-term relapse of SNB angle between T2-T3 was not significant, however, SNB angle was reduced at T3 (SNB angle, mean 75.5°; range 70.1 to 83.1°), and long-term advancements between T1-T3 were not significant. BSSO procedures were performed in 13 patients, and genioplasty was performed in 9 patients.

No significant changes were observed in occlusal plane inclination nor in anterior facial height. The T2 values on anterior facial height were not included. For details see Table 4 and Figure 7 and 8.

Mandibular lengthening was observed postoperatively at T1-T2 from mean 79.7 mm (range 62.4 to 103.6 mm) to 87.2 mm (range 75.3 to 113 mm); $P_{T1/T2}$ = 0.004, and lengthening was stable at long-term, however, not significant (mean 89.8 mm; range 72.4 to 113.1 mm; $P_{T1/T3}$ = 0.056) at T3. Details provided in Table 4 and Figure 8.

HOB improved significantly from mean 5.3 mm (range 1.8 to 9.2 mm) at T1 to 2.7 mm (range 1.3 to 5.4 mm) at T3 ($P_{T1/T3}$ = 0.004). Preoperative open bite was closed at long-term (VOB, mean -0.7 to 1.5 mm), however, changes were not significant. T2 values on HOB and VOB were not included. Details shown in Table 4 and Figure 5.

No significant effect was observed on ramus height ratio or chin deviation.

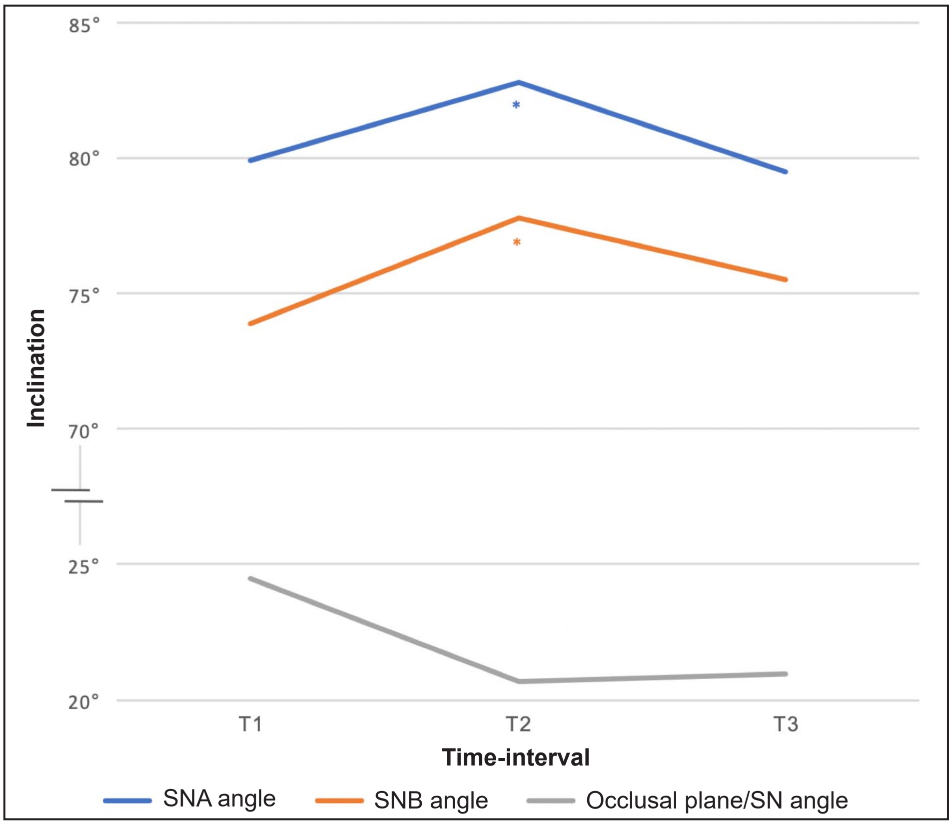

Figure 7. Jaw advancements and occlusal plane inclination.
T1 = preoperative examination; T2 = postoperative examination; T3 = long-term examination.
*$P_{T1/T2} < 0.05$, paired-samples t-test for differences in SNA and SNB angle within groups.

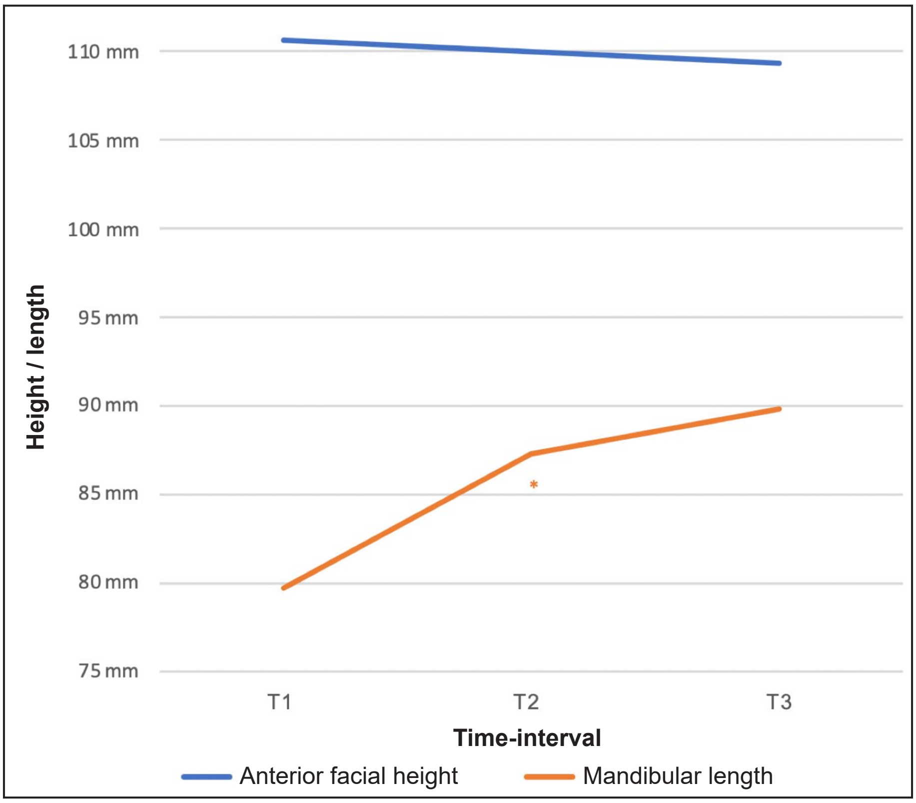

Figure 8. Anterior facial height and mandibular length.
T1 = preoperative examination; T2 = postoperative examination; T3 = long-term examination. T2 values on anterior facial height were not included.
*$P_{T1/T2} < 0.05$, paired-samples t-test for differences in mandibular length within groups.

DISCUSSION

This retrospective two-center study evaluated the outcomes of OS treatment of dentofacial deformities caused by JIA involving the TMJ. As shown in a recent systematic review, studies on OS in this patient group suffer from small sample size and short follow-up [19]. We report on 19 patients with a mean follow-up on orofacial symptoms/clinical findings and radiologic findings of 3.8 and 2.6 years, respectively.

Patients who underwent orthognathic surgical treatment experienced stable normalisation of incisal relations and stable mandibular lengthening with no negative long-term effects on TMJ function or orofacial symptoms.

Patients included were highly heterogeneous in terms of disease, affection and individual treatment course. This variance was accepted due to sample size, but has to be considered when concluding from the results. Several issues are to be considered during treatment planning, e.g. function of the arthritis affected TMJ, stability of the mandibular condyle and disease remission. No evidence exists concerning the optimal combination of surgical procedures in relation to these issues.

Although craniofacial pain and dysfunction have been reported to increase during the natural course of the disease in JIA patients [10], we did not detect a worsening of orofacial symptoms recorded during the 45.8-month follow-up period based on available data from 14 to 17 patients. Except from a temporary and insignificant decrease in jaw mobility postoperatively, no significant changes were found in TMJ function and jaw mobility.

Observations on orofacial symptoms and TMJ function were generally in line with previous studies on the effect of OS in patients with JIA and TMJ involvement, showing no [25-27] or minimal [28] negative effect on orofacial symptoms and minimal long-term effect on TMJ function [29].

Facial symmetry was evaluated clinically, i.e. facial asymmetry and chin deviation, and radiologically, i.e. ramus height ratio and chin deviation. Short- and long-term changes in facial symmetry were not statistically significant, however, a tendency was observed towards improved clinical facial symmetry and chin deviation at long-term (35 and 36% reduction, respectively, $P_{T1/T3}$ = 0.055 in each case). Although soft tissue asymmetry generally follows skeletal asymmetry [30], no radiologic effect was observed on posterior facial symmetry or chin asymmetry. A possible explanation might include that a threshold for assessing differences at radiologic landmarks might not be numerical differences significant [30].

Clinical registrations on HOB and VOB indicate stable closure of anterior open bite with normalisation of vertical and horizontal incisal relations. Although radiographic measurements were not significant regarding reduction of VOB, a positive vertical overlap was found. A recent study reported similar results with improved and stable HOB and VOB following corrective jaw surgery in 11 of 12 patients with JIA [29].

Results indicate significant postoperative bimaxillary sagittal advancements between T1-T2 (SNA, mean 2.9° advancement, $P_{T1/T2}$ = 0.022; SNB, mean 3.9° advancement, $P_{T1/T2}$ = 0.003) with improved and stable mandibular lengthening. Long-term observations indicate a relapse of the gained maxillary advancement as well as some mandibular relapse. Relapse was surprising considering the normal sagittal relations between both jaws and the cranial base at T3 and the stable normalisation of incisal relations. It may be speculated whether observed maxillary relapse was affected by segmental Le Fort I procedures (rotation of the anterior segment with surgical retraction of incisors and advancement of the A point), postoperative orthodontics, or perhaps the analysts were unable to detect changes within the relatively small advancements.

Previous studies on stability after OS in a comparable patient group have reported different outcomes. No skeletal relapse was reported by both Raffaini et al. [31] (13 patients; all bimaxillary surgery and 11 with additional genioplasty; measured at pogonion) and Pagnoni et al. [27] (5 patients; all Le Fort I and genioplasty; measured at A point/B point/pogonion), whereas Oye et al. [28] (16 patients; 12 genioplasty, 10 BSSO, 1 Le Fort I; measured at pogonion) and Stoor et al. [29] (12 patients; combinations of OS, MDO and/or TMJ prosthesis; measured at A point/B point/mandibular length) reported significant and moderate-to-minor relapse, respectively.

Skeletal relapse may be due to several factors, i.e. greater advancement and greater reduction in mandibular plane angle may have played a role in horizontal and vertical relapse [32-36]. Additionally, existing TMJ deformity/dysfunction, potential disease activity, young age, type of osteosynthesis of the BSSO and proximal segment control are all variables that are known to affect the stability of OS [37-40].

The strengths of this retrospective two-center study were the standardised examination and data collection as well as comprehensive datasets and calibration of surgeons.

Limitations of the study include a retrospective study design with relatively small sample size and no uniform range of follow-up. Patient- and treatment characteristics were not homogenous, although, was accepted due to population size and follow-up, and was the result of the highly individual disease- and treatment course of these patients. No intra- or inter-rater reliability testing was performed, and datasets were not entirely complete.

CONCLUSIONS

Treatment with combinations of orthognathic surgical treatment including bilateral sagittal split osteotomy, Le Fort I and/or genioplasty with orthodontics in patients with temporomandibular joint juvenile idiopathic arthritis and dentofacial deformities provided stable normalisation of incisal relations and mandibular lengthening without deterioration of long-term temporomandibular joint function or orofacial symptoms, and without activation of disease. Postoperative jaw advancements were indicated, but no significant effect was evident at long-term. Also, a trend towards improved clinical facial symmetry and chin deviation was observed at long-term.

We found a need for future prospective studies focusing on surgical procedures in order to optimize treatment outcome in patients with temporomandibular joint juvenile idiopathic arthritis and dentofacial deformities.

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