

Orthodontic treatment of a particular subgroup of children with special health care needs, children with craniofacial anomalies:

An analysis of treatment length and clinical outcome

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ABSTRACT

Objective: To analyze any differences in the orthodontic treatment between children belonging to a particular subgroup of subjects with special health care needs (SHCN), children with craniofacial anomalies (CFA), and children not diagnosed with SHCN (NO SHCN).

Materials and Methods: The study sample consisted of 50 children with SHCN and a confirmed diagnosis of CFA (SHCN/CFA); the control group consisted of 50 NO SHCN children fully matched for age, gender, and type of appliance used. The differences between the two groups were analyzed retrospectively: pre-, posttreatment scores, and score reduction of the Peer Assessment Rating Index (PAR), dental health component (DHC), and aesthetic component (AC) of Orthodontic Treatment Need Index (IOTN), number of appointments, number of simple or complex chair-time appointments, overall treatment time, and age at treatment start and end.

Results: There were no statistically significant differences between the SHCN/CFA and NO SHCN groups for number of appointments, overall treatment time, age at treatment start, and age at treatment end ($P = .682, .458, .535, \text{ and } .675$, respectively). There were statistically significant differences between groups in PAR, DHC, AC pre- and posttreatment, and number of simple and complex chair-time appointments ($P = .030 \text{ and } .000; .020 \text{ and } .023; .000 \text{ and } .000; .043; \text{ and } .037$; respectively). The reduction of PAR, DHC, and AC scores was not significantly different between groups ($P = .060, .765, \text{ and } .825$, respectively).

Conclusion: The treatment of children with SHCN, in general, and with CFA, in particular, on the one hand involves a higher rate of using complex chair time appointments and an inferior treatment outcome, by the other side implies an overall treatment time and a reduction of PAR, DHC or AC scores similar to the treatment of children not diagnosed with SHCN. (*Angle Orthod.* 2016;86:115–120.)

KEY WORDS: Children with special health care needs; Children with craniofacial anomalies; Peer Assessment Rating; Index of Orthodontic Treatment Need; Dental health component; Aesthetic component

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INTRODUCTION

According to the WHO definition and the craniofacial imaging experience of the St. Louis Children's Hospital, craniofacial anomalies (CFAs) are congenital structural deformities, malformations, or other abnormalities of the skull or facial bones, which appear to arise from a combination of genetic factors and environmental influences.¹ The term *SHCN* was defined according to the international classification of functioning, disability, and health.²

Advances in medical treatment for newborns with developmental and/or environmental restrictions have led to significant increases in survival chances; consequently, the number of children with special health care needs (SHCN) continues to rise.³ The integration of subjects with special needs and their

families into mainstream daily and social life is becoming increasingly important.⁴

Children having CFAs are a subgroup of a wide spectrum of patients with SHCN that shares a unique facial appearance that exposes them to society.⁵ People with normal dental appearance are perceived to be better looking, more desirable to make friends with, more intelligent, and less likely to show aggressive behavior.⁶ Facial and dental appearance make a difference in an individual's social integration. The primary motivation for parents to have their SHCN children undergo orthodontic therapy is to improve their facial attractiveness.^{4,7} This information and the fact that malocclusions occur more often in subjects with SHCN suggest that access to orthodontic treatment for patients with CFAs is strongly recommended to improve their quality of life.⁸ Current literature suggests that orthodontic treatment in children with SHCN is possible, but data are scarce regarding their management and orthodontic therapy outcomes.⁹

The use of standardized indices facilitates evaluation of malocclusion and esthetics, often considered as subjective criteria and difficult to evaluate; a manifold number of indices for orthodontic assessment have been described in the literature.¹⁰ The two most commonly used indices are the Peer Assessment Rating (PAR) Index and the Index of Orthodontic Treatment Need (IOTN),¹¹ which were applied in our study in order to examine differences between the SHCN/CFA and the not-diagnosed-with-SHCN (NO SHCN) groups.

The PAR Index provides an estimate of how far a case deviates from normal alignment and occlusion: 0 indicates perfect alignment and occlusion while higher scores (rarely beyond 50) indicate increasing levels of irregularity. The difference in scores between the pre- and posttreatment cases reflects the degree of improvement and, therefore, the success of treatment.^{12,13}

The IOTN is composed of both a dental health component (DHC) and an aesthetic component (AC).¹¹ The five grades within the DHC have been grouped as follows: grade 1 = no treatment required, grade 2 = little treatment required, grade 3 = moderate or borderline treatment required, grade 4 = great or severe need of treatment required, and grade 5 = very great need of orthodontic treatment required.¹⁴

The AC consists of a scale of ten photographs showing different levels of dental attractiveness, grade 1 representing the most attractive and grade 10, the least attractive dentitions. Grades 1–4 represent little or no treatment required, grades 5–7 represent moderate or borderline treatment required, and grades 8–10 represent the strongest indication for treatment.¹⁵ Although Hunt et al. suggested that the first category be grades 1–3 rather than 1–4, the AC would at least

help identify potentially cooperative patients who are also interested in treatment and should, therefore, be considered a reliable method of assessing the subjective orthodontic treatment need.^{15,16}

Our intention was to explore the subject related to the orthodontic therapy of patients with SHCN in general and with CFAs in particular, considering that there is little in the literature of that nature.^{4,7–9,17–21} The aim of this study was to analyze treatment time and differences between pre- and posttreatment PAR and DHC, and AC scores of IOTN, in a SHCN/CFA group compared with a NO SHCN control group.

MATERIALS AND METHODS

Study and Control Group Selection

The study sample consisted of 50 orthodontically treated children with SHCN and an SHCN/CFA (36 males and 14 females; mean age at treatment start, 9 ± 1.0 and at treatment end, 13 ± 0.7 years). The sample included several conditions such as Down syndrome ($n = 36$),²² Goldenhar syndrome ($n = 2$),²³ Treacher Collins-Franceschetti syndrome ($n = 2$),²⁴ Gorlin-Goltz syndrome ($n = 2$),²⁵ Apert-Crouzon syndrome ($n = 1$),²⁶ and cleft lip-palate ($n = 7$),²⁷ all summarized in Table 1. The control group consisted of 50 orthodontically treated NO SHCN children (36 males and 14 females; mean age at treatment start, 9 ± 1.0 and at treatment end, 12.8 ± 0.7). Study and control groups were matched for age, gender, and type of appliance used (36 patients were treated with fixed and removable appliances, 10 were treated with fixed appliances only, and 4 with removable appliances only).

Inclusion criteria of the SHCN/CFA group were confirmed diagnosis of a CFA, orthodontic nonsurgical therapy using removable and/or fixed appliances, and availability of photographic and model documentation at the beginning and end of treatment. Exclusion criteria were patients with a previous history of orthodontic therapy, incomplete medical records, or discontinuation of treatment. Inclusion and exclusion criteria for the NO SHCN group were identical to those for the SHCN/CFA group, except for the confirmed diagnosis of a CFA.

The subjects included in this study were patients attending the Department of Biomedical-Neuromotor Sciences, Dentistry for Special Needs Patients Division, Dental School, University of Bologna, Italy. The study protocol and informed written consents were obtained in full accordance with the ethical principles of the Declaration of Helsinki of 1975 and approved by an institutional review board and by the local ethics committee of the university (PG. N 0019293 20/06/2014).

Table 1. Description of the Study Sample^a

SHCN/CFA	N			Age at Start of Treatment			Age at End of Treatment			Type of Appliance Used		
	N	Male	Female	Mean ± SD	Min	Max	Mean ± SD	Min	Max	Removable	Fixed	Removable and Fixed
Down syndrome	36	23	13	9 ± 1.5	3	10	13 ± 0.7	11	14	4	8	24
Goldenhar syndrome	2	2	0	9.5 ± 0.7	9	10	13.5 ± 0.7	13	14	0	0	2
Treacher Collins-Franceschetti syndrome	2	2	0	9.5 ± 0.7	9	10	13.5 ± 0.7	13	14	0	0	2
Gorlin-Goltz syndrome	2	2	0	9.5 ± 0.7	9	10	13.5 ± 0.7	13	14	0	0	2
Apert-Crouzon syndrome	1	1	0	9.5	9.5	9.5	13.5	13.5	13.5	0	0	1
Cleft lip-palate	7	6	1	8.5 ± 0.3	8	9	12.5 ± 0.7	11	13	0	2	5
Total number	50	36	14	9 ± 1.0	3	10	13 ± 0.7	11	14	4	10	36

^a Range (R) for the age at treatment start of each of the diagnoses mentioned: Down syndrome, R = 7; Goldenhar syndrome, R = 1; Treacher Collins-Franceschetti syndrome, R = 1; Gorlin-Goltz syndrome, R = 1; Apert-Crouzon syndrome, R = 0; Cleft lip-palate, R = 1. Range for the age at end of treatment of each of the diagnoses mentioned: Down syndrome, R = 3; Goldenhar syndrome, R = 1; Treacher Collins-Franceschetti syndrome, R = 1; Gorlin-Goltz syndrome, R = 1; Apert-Crouzon syndrome, R = 0; Cleft lip-palate, R = 2.

Duration of Each Treatment Session: Simple or Complex Chair Time

The dates of the start and completion of orthodontic treatment were registered. The date of birth was also identified and used to compare patients' ages at the first and last appointments. Furthermore, the medical disorder, gender, type of appliance used (fixed or removable), and number of appointments subdivided on the basis of chair time as either simple or complex were recorded. Because of the retrospective nature of the study we have divided, according to the duration of each treatment session, the appointments of all patients into simple or complex chair time. Therefore, we defined simple chair time as less time-consuming orthodontic treatments such as changing elastics or power chains and routine adjustment of removable appliances, whereas complex chair time implied procedures such as wire change, bracket bonding or rebonding, dental impressions, and first adjustment of a removable appliance.

Peer Assessment Rating

The PAR was used to examine improvement in occlusion between pre- and posttreatment and to compare the overall treatment outcome between the two groups.

The examination team consisted of the first author (a trained and qualified orthodontist in the use of PAR and IOTN) and a postgraduate dental student who acted as a recorder.

The PAR Index components were recorded and summed for all pre- and posttreatment dental study models with the aid of a PAR ruler. The 11 components of PAR evaluated were maxillary right segment, maxillary anterior segment, maxillary left segment, mandibular right segment, mandibular anterior segment, mandibular left segment, right buccal occlusion, left buccal occlusion, overjet, overbite, and midline.¹²

Dental Health Component of the IOTN

The DHC of the IOTN was recorded to assess the change in the patient's orthodontic treatment need over the entire duration of therapy.¹⁴ The Community Periodontal Index probe, a metallic millimeter ruler, and a mouth mirror were used for data collection by the examiner; the DHC variables containing pre- and posttreatment values were recorded and tabulated individually for each child.

Aesthetic Component of the IOTN

The AC of the IOTN was recorded to assess the change in the patient's dental attractiveness over the entire treatment time, requiring the examiner to compare the patient's frontal intraoral photographs with 10 standardized photographs that ranged from 1, for the most attractive, to 10, for the least attractive dental arrangement.¹⁵ Therefore, the Standardized Continuum of Aesthetic Need Index was used to rate the patient's frontal intraoral photographs on a visual analog scale.

Statistical Analysis

Weighted (Fleiss-Cohen) kappa statistics for the clinician were used to confirm the examiner calibration and to assess intra-examiner reliability, which was measured by rescoring a random sample of 10 models and photographs, for the study and control groups, 6 weeks apart.

The data were entered in MS Excel and analyzed using SPSS version 21.0 statistical software (IBM Corp, Armonk, NY). The Mann-Whitney *U* test was performed to compare the number of appointments, number of simple and complex chair time appointments, overall treatment time (defined as the time interval between the start and end of active treatment),

Table 2. Descriptive Statistics of the Collected Data^a

Collected Data	Total Sample			SHCN/CFA			NO SHCN			SHCN/CFA
	N = 100			n = 50			n = 50			Vs. NO SHCN
	Median	Min	Max	Median	Min	Max	Median	Min	Max	<i>P</i> Values
PAR										
Pretreatment	18	6	45	20	10	45	16	6	31	.030*
Posttreatment	3	0	40	8	2	40	0	0	9	<.001*
Score reduction	13.5	-1	32	12	-1	32	15	3	29	.06
DHC										
Pretreatment	4	2	5	4.5	4	5	3.5	3	4	.020*
Posttreatment	2.5	1	4	3	2	4	2	1	3	.023*
Score reduction	1.5	1	3	1.5	1	3	1.5	1	3	.765
AC										
Pretreatment	7	1	10	9	5	10	5	1	10	<.001*
Posttreatment	2	1	10	3	1	10	1	1	5	<.001*
Score reduction	4	0	9	4	0	9	4	0	8	.825
Number of										
Appointments	24.5	12	54	26	15	54	23	12	46	.682
Simple chair time appointments	13	5	33	10	5	22	16	8	33	.043*
Complex chair time appointments	10	4	32	15	10	32	6	4	13	.037*
Treatment length (mo)										
Overall time of orthodontic treatment (mo)	34	12	96	48	18	108	46	14	96	.458
Age (y)										
	Mean ± SD	Min	Max	Mean ± SD	Min	Max	Mean ± SD	Min	Max	
At treatment start	9 ± 1.0	3	10	9 ± 1.0	3	10	9 ± 1.0	4	10	.535
At treatment end	12.9 ± 0.7	11	14	13 ± 0.7	11	14	12.8 ± 0.7	11	13.5	.675

^a The Mann-Whitney *U* test was performed to compare between SHCN/CFA and NO SHCN groups.

* Values considered significant at <.05.

reduction in PAR scores (difference between the pre- and posttreatment PAR scores), reduction in DHC scores (difference between the pre- and posttreatment DHC scores), and reduction in AC scores (difference between the pre- and posttreatment AC scores) between the SHCN/CFA and NO SHCN groups.

RESULTS

The collected data and results of the statistical analysis are shown in Table 2. There were no statistically significant differences in the overall treatment time or number of appointments between the two groups ($P = .458$ and $.682$, respectively). The SHCN/CFA group needed more complex and less simple chair time appointments compared with the NO SHCN group; the difference was statistically significant ($P = .037$ and $.043$, respectively).

Mean age of the patients was not statistically different, between groups, at the time of treatment start (mean age, 9 ± 1.0 years for the SHCN/CFA and NO SHCN groups), or treatment end (mean age, 13 ± 0.7 years for the SHCN/CFA group and 12.8 ± 0.7 for the NO SHCN group) ($P = .535$ and $.675$, respectively).

The PAR Index pre- and posttreatment was significantly higher in the SHCN/CFA group than in the NO

SHCN group ($P = .030$ and $<.001$, respectively). Reduction in the PAR score (pre- and posttreatment) was not statistically significant between the two groups ($P = .060$).

There was a significant difference in pre- and posttreatment DHC values between the SHCN/CFA group and the NO SHCN group ($P = .020$ and $.23$, respectively). The DHC score reduction was not statistically different between the study and control groups ($P = .765$).

The aesthetic component showed significant differences at treatment start and end between the SHCN/CFA and NO SHCN groups ($P < .001$ for both the AC pre- and posttreatment). Statistically significant differences were not present for the AC score reduction between the two groups ($P = .825$).

DISCUSSION

The results of this study showed that overall treatment time in SHCN/CFA children and in NO SHCN children were not statistically different. The same findings applied to the total number of appointments. The study group showed a higher number of complex chair time appointments; the control group showed a higher number of simple chair time appointments.

These differences could be explained by the cognitive and physical limitations of children with SHCN, which led to more complications with orthodontic appliances.

The age of patients in both groups at the start and end of treatment was not statistically different. In both groups, the minimum age at the beginning of therapy was very early. In 50 patients with SHCN/CFA, treatment was begun early in two subjects with Down syndrome because of using Castillo-Morales stimulating plates.²⁸ In 50 NO SHCN children, 2 were treated early with space maintainers according to the E-space preservation technique.²⁹

Differences in the PAR, DHC, and AC scores between the SHCN/CFA and NO SHCN groups, to the disadvantage of the first group, could be detected. This observation was explained by Townsend and coworkers, who identified genetic and environmental factors that influenced dentofacial morphology.³⁰ Environmental factors such as generalized muscular hypotonia were often found in patients with SHCN. Inferior outcomes of orthodontic treatment, therefore, could be expected in this particular group, as indeed it was found to be. These results may be attributed to the fact that parents and orthodontists of SHCN children focus more on functional outcome and are willing to subordinate aesthetic effects. In addition, inadequate conditions of oral hygiene may force the orthodontist to reduce the treatment time in order to avoid dental damage.

Differences in the reduction of the PAR, DHC, and AC scores were not found between the two groups, showing a significant reduction in the scores. These findings confirm the influence of the genetic predisposition of the subjects on the treatment outcome.³⁰

The selection of patients with CFAs, a subgroup of a wide spectrum of patients with SHCN, forms a heterogeneous sample group with different characteristics. This study focused on children with CFAs because it is precisely this group, within the patients with SHCN, that is more often affected by malocclusions and probably well represents SHCN subjects in the daily orthodontic practice.^{7,8,17} On the other side, since one of the inclusion criteria for CFA patients was having had nonsurgical orthodontic treatment, the more severe clinical cases were excluded from the study group. At the same time, since there were no inclusion or exclusion criteria for children with CFAs regarding the levels of cooperation and oral hygiene, the CFA subjects should be more representative of the patients with SHCN. The indexes applied have been well established, standardized, scientifically examined, and compared with each other.^{10–16} As shown by Brown and Inglehart,²⁰ there has to be a reason why orthodontists hesitate to provide care for children with SHCN. It is important to encourage a further

investment of economic resources and funds for orthodontic treatment of patients with SHCN in order to influence daily practice and, therefore, improve these subjects' quality of life.

Further prospective research is encouraged with a larger sample and specific inclusion criteria as to type of malocclusion, its severity, and appliances implemented. It is hoped that this study will encourage orthodontists to implement the treatment of children with SHCN in general and with CFAs in particular to promote all care options and make therapy accessible to these subjects.

CONCLUSIONS

- The overall treatment time and number of appointments showed no statistically significant differences between the SHCN/CFA and NO SHCN groups.
- Children with a SHCN/CFA had a higher rate of using complex chair time appointments compared with the NO SHCN patients.
- An inferior treatment outcome rated by PAR, DHC, and AC scores for children with SHCN/CFAs was observed.
- No statistically significant differences in the reduction of PAR, DHC, or AC scores were found between groups.

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