Does Hyoid Bone Resection According to Sistrunk Influence Normal Craniofacial Growth? A Cephalometric Study

Isabella M. Joss-Vassalli, DDS, Christof U. Joss, DDS, MSc,† and Urs Gebauer, DDS‡*

Purpose: To retrospectively evaluate the influence of hyoid bone resection according to Sistrunk in early age due to a thyroglossal duct cyst on craniofacial growth.

Materials and Methods: We retrospectively examined 10 patients (2 females and 8 males) having had hyoid bone resection according to Sistrunk due to thyroglossal duct cysts by lateral cephalograms taken before orthodontic treatment (mean, 17.1 years; range, 8.6-31.9 years). Surgery was carried out at a mean age of 4.4 years (range, 0.37-9.8 years). All lateral cephalograms were evaluated and traced by hand. Descriptive statistics were calculated, and data from each patient were compared individually with corresponding standard values (age and gender) from Bathia and Leighton.

Results: With regard to sagittal parameters, the SNB angles were by trend too small and the ANB angles were too large. However, the ratio of mandibular to maxillary length showed that the patients had a mandible that was too large or maxilla that was too small. With regard to vertical parameters, large deviations from normal values in both directions (hyperdivergent to hypodivergent pattern) could be detected when we analyzed NSL/ML′, NL/ML′, and NSL/NL. With regard to dental parameters, the majority of the patients had retroclined upper (IsL/NL, IsL/N-A) and lower (IiL/ML, IiL/N-B) incisors.

Conclusions: Several vertical and horizontal skeletal and dental cephalometric parameters were shown to be different by trend when compared with control values. A possible negative impact on craniofacial growth potential and direction as a result of hyoid resection in early age according to Sistrunk cannot be excluded.

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The tongue position is believed to have a major impact on the development of the maxilla and mandible. The muscles of the tongue and the suprahyoid muscles have their insertions at the hyoid bone. The interaction between growth direction of the mandible, tongue position, and hyoid position has been the subject of different research groups and is still not clearly understood.[1-4]

The main idea of this study was to have the possibility to examine patients who had lost their hyoid bone in some way in early childhood and to analyze its influence on craniofacial growth. Patients with median neck cysts or so-called thyroglossal duct cysts (TGDCs) have their hyoid bone partly resected to prevent reappearance of TGDCs.[5] To our knowledge, no reports on the influence of hyoid bone resection on craniofacial growth have been published in the literature. Furthermore, it is not known whether the method of Sistrunk[5] has any negative impact on normal craniofacial growth and development.

TGDCs are rare congenital malformations of the thyroid apparatus, marked by swelling and inflammation of the anterior neck. However, rare carcinomas arising from TGDCs have been described.[6-8] TGDCs are the most common form of congenital neck cysts, accounting for up to 70% of such lesions. They are commonly present in children and adolescents, but in up to one third of patients it is only detected at 20

*Member of Senior Staff, Department of Orthodontics and Dentofacial Orthopedics, University of Berne, Berne, Switzerland.

†Research Assistant, Department of Orthodontics, University of Geneva, Geneva, Switzerland; and PhD Student, Department of Orthodontics and Oral Biology, Radboud University Nijmegen Medical Center, Nijmegen, The Netherlands.

‡Formerly, Professor and Head, Department of Orthodontics and Dentofacial Orthopedics, University of Berne, Berne, Switzerland.

Address correspondence and reprint requests to Dr Joss-Vassalli: Klinik für Kieferorthopädie, Zahnmedizinische Kliniken der Universität Bern, Freiburgstrasse 7, CH-3010 Berne, Switzerland; e-mail: isabella.vassalli@zmk.unibe.ch

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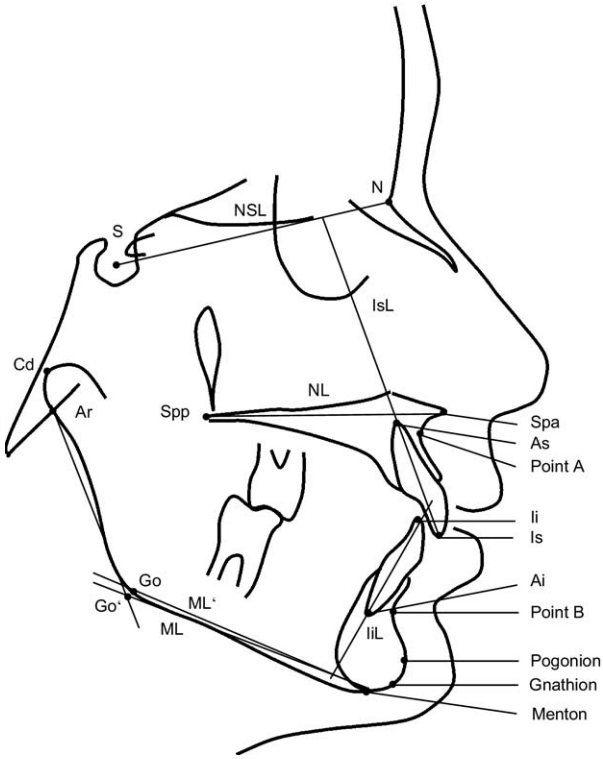

FIGURE 1. Reference points and lines used in cephalometric analysis. S, Sella; NSL, nasion-Sella line; N, nasion; IsL, upper incisal line; Cd, condylion; Ar, articulare; NL, nasal line; Spp, posterior nasal spine; Spa, anterior nasal spine; As, upper incisor apex; IiL, lower incisal line; Go, gonion; Go′, tangent gonion; ML, mandibular line; ML′, mandibular line′; Ii, incision inferior; Is, incision superior; Ai, lower incisor apex.

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years or older. Male patients and female patients are equally affected.[9] A meticulous clinical history and physical examination are sufficient to make the diagnosis in most cases. Simple cystectomies without hyoid bone removal according to Sistrunk[5] often lead to recurrence of the TGDC and reduce the chance of healing after a second correctly undertaken operation with hyoid resection. Therefore the method of Sistrunk remains the therapy of choice.[10]

Ellis and van Nostrand[11] found the incidence of TGDCs in a postmortem study of 200 adults to be as high as 7%. The inheritance pattern is usually autosomal dominant with incomplete penetrance, and a female preponderance has been noted.[12]

The aim of this study was to evaluate retrospectively the influence of hyoid bone resection according to Sistrunk[5] in early age due to a TGDC on craniofacial growth.

Materials and Methods

Hyoid bone resection was performed in 40 patients (28 males and 12 females) in early childhood due to the presence of a median neck cyst or so-called TGDC at the Departments of Pediatric Surgery and Maxillofacial Surgery,[10] Inselspital, University of Berne, Berne, Switzerland. The medial part of the hyoid bone, the cyst, and fistulas, if present, underwent monoblock excision according to the method of Sistrunk.[5] The surgical technique for the resection of the hyoid bone was the same for all patients, and each of the surgeons was experienced in this procedure. All patients were operated on in 1984 to 1994. To analyze the influence on craniofacial growth, it was crucial to exclude patients in whom TGDC surgery was done too late in development (ie, late childhood or adulthood).

Twenty-four patients could be located. The data of 10 patients (2 female and 8 male patients, all of whom were white), aged 8.6 to 31.9 years (mean age, 17.1 years), could be analyzed in this study. In patients who had undergone orthodontic therapy, initial lateral cephalograms were requested from their orthodontists to exclude the influence of orthodontic therapy on craniofacial growth. In the other patients, lateral cephalograms were obtained at the Department of Orthodontics, University of Berne. Ethical approval was obtained from the ethics committee of Canton Berne, Switzerland (No. 48/05). All subjects provided written, informed consent.

The cephalometric analysis and tracings were carried out by 1 author (I.M.J.-V.) and included the ref-

Table 1. ACCIDENTAL ERRORS OF CEPHALOMETRIC ANALYSIS

Variable	Accidental Error
SNA (°)	0.57
SNB (°)	0.34
ANB (°)	0.46
NSL/NL (°)	0.66
NSL/ML′ (°)	0.42
NL/ML′ (°)	0.63
N-S-Gn (°)	0.45
IsL/IiL (°)	1.22
IsL/NL (°)	0.81
IsL–N-A (°)	0.83
IiL–N-B (°)	1.00
IiL/ML′ (°)	1.10
Anterior lower face height (mm)	0.46
Anterior total face height (mm)	0.46
Posterior total face height (mm)	0.65
Anterior lower face height/posterior total face height	0.00
Anterior total face height/posterior total face height	0.01
Cd-Pg (mm)	1.14
Cd-Spa (mm)	0.96
Mandibular length/maxillary length	0.01

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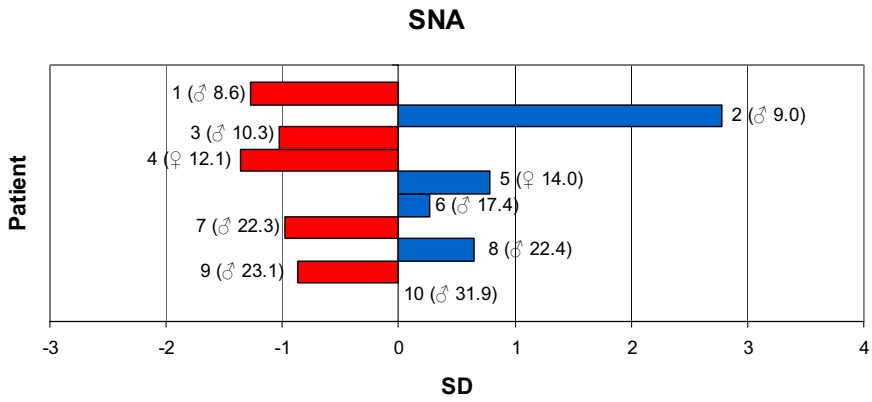

FIGURE 2. SNA angle. Patient number, gender, and age are indicated next to each bar.

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erence points and lines shown in Figure 1. Anterior total face height is the measured sum of the lines from nasion perpendicular to NL and menton perpendicular to NL. Anterior lower face height is the length of the line from menton perpendicular to NL. Posterior total face height is the distance between Sella and gonion perpendicular to NL. Maxillary length is calculated from condylion to Spa and mandibular length from condylion to pogonion.

ERROR OF METHOD

The systematic and accidental errors of the cephalometric analysis were evaluated by duplicate determinations of all 10 cephalograms. The cephalograms were retraced and remeasured for a second time by the same author 2 weeks after the first assessment. No systematic errors were found when the values were evaluated with a paired *t* test. The accidental errors (*si*) were calculated with the following formula:

$$si = \sqrt{\frac{\sum d^2}{2n}}$$

where *d* is the difference between the repeated measurements and *n* is the number of duplicate determinations.[13] These errors are shown in Table 1.

Most of the angular variables and coordinates of the skeletal reference points had accidental errors smaller than 1.0° and 1.0 mm, respectively. The exceptions were IsL/IiL (1.22°), IiL/ML′ (1.10°), and Cd-Pg (1.14 mm).

STATISTICAL METHODS

For the standard cephalometric analyses, the growth standards of Bathia and Leighton[14] were used as control parameters. Each patient was allocated according to his or her age when the lateral cephalogram was obtained and considering gender differences. Bathia and Leighton only followed their subjects up to 20 years of age. For the 4 patients who were aged 22.3, 22.4, 23.2, and 32.0 years, the control parameters of 20-year-old patients were used.

Given the relative rarity of TGDCs, heterogeneity of age and a lack of female patients were present in the analyzed sample of 10 patients. Furthermore, differences in age and gender affect the developmental stage in craniofacial growth of the patient to a large

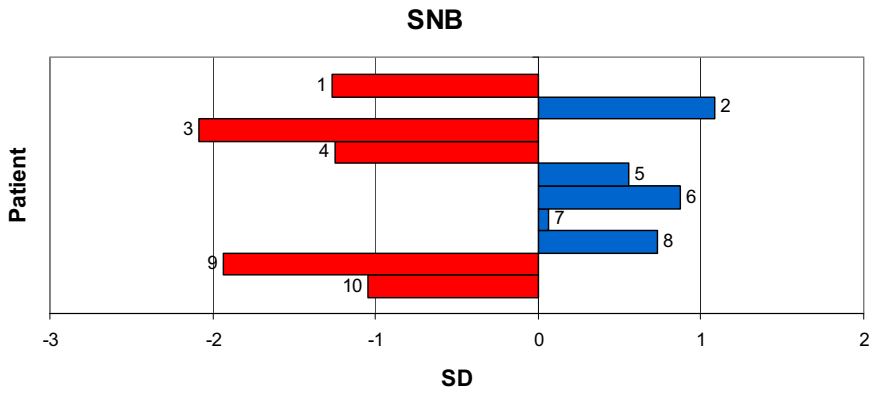

FIGURE 3. SNB angle.

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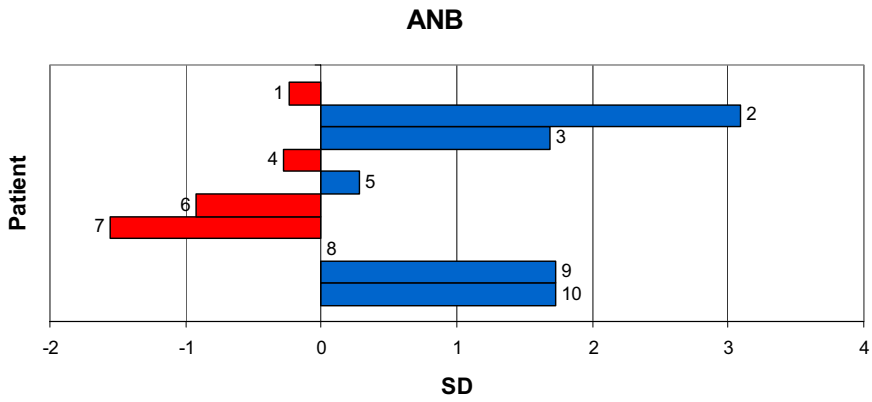

FIGURE 4. ANB angle.

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extent. For this reason, each patient was analyzed and compared separately according to the referring control group. Because of partially unknown enlargement factors for lateral cephalograms obtained in some orthodontic offices, only angles and ratios but no linear variables were included in the cephalometric analysis.

Descriptive statistics were chosen for each angular variable or calculated ratio. The values were relocated in the distribution described by Bathia and Leighton.[14]

Results

SKELETAL PARAMETERS

Sagittal Dimension

Six patients have SNB angles outside the first SD compared with the Bathia and Leighton group.[14] Five of them have an SNB angle that is too small. Regarding the ANB angle, 4 of 5 patients, whose ANB angles are outside the first SD, have an ANB angle that is too large. However, a tendency toward larger ANB angles due to smaller SNB and normal SNA angles can be observed (Figs 2-4).

In 70% of all patients, the ratio of maxillary length to mandibular length is above the first SD when compared with the Bathia and Leighton group[14] (Fig 5). There is a trend in this patient sample of having a maxilla that is too small or mandible that is too large.

Vertical Dimension

A more heterogeneous pattern with large deviation in both directions is seen in the NSL/ML′ and NL/ML′ angles compared with normal values. The NSL/ML′ angle was outside the first SD in 50% of all patients and the NL/ML′ angle was outside the first SD in 40% of all patients when compared with the Bathia and Leighton group.[14] The NSL/ML′ angle was even outside the second SD in 40%, and the NL/ML′ angle was outside the second SD in 30%. In half of all patients whose NSL/ML′ angles were outside the first SD, the

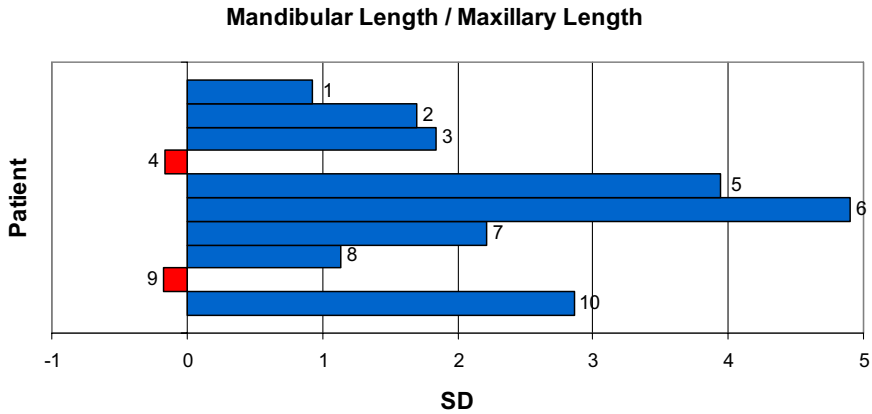

FIGURE 5. Ratio of mandibular length to maxillary length.

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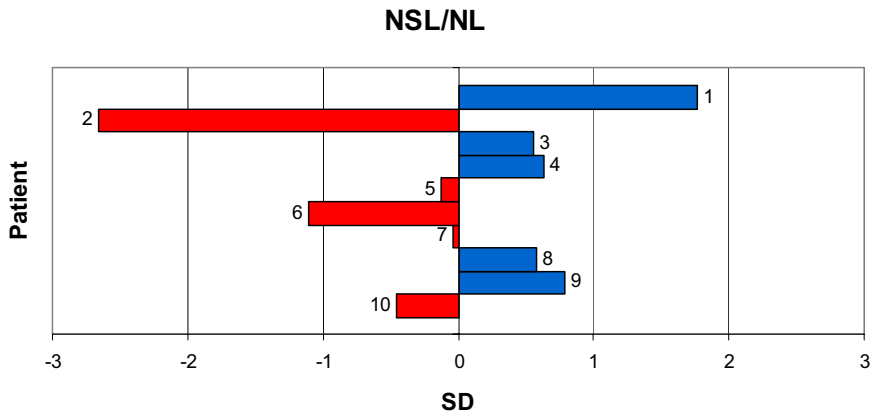

FIGURE 6. NSL/NL angle.

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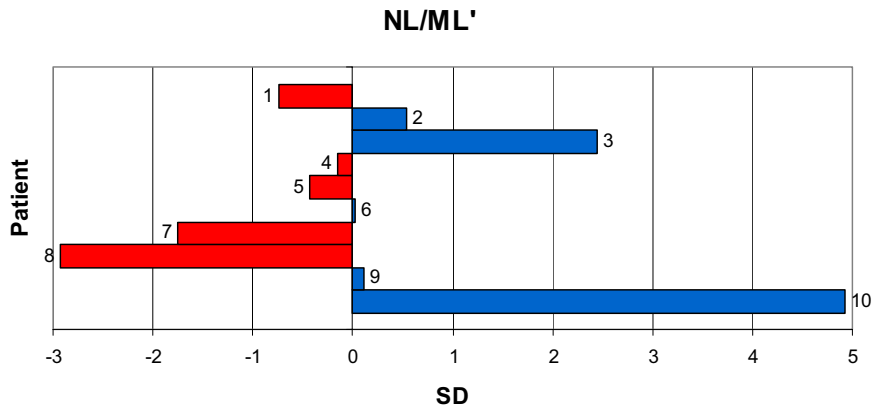

FIGURE 7. NL/ML′ angle.

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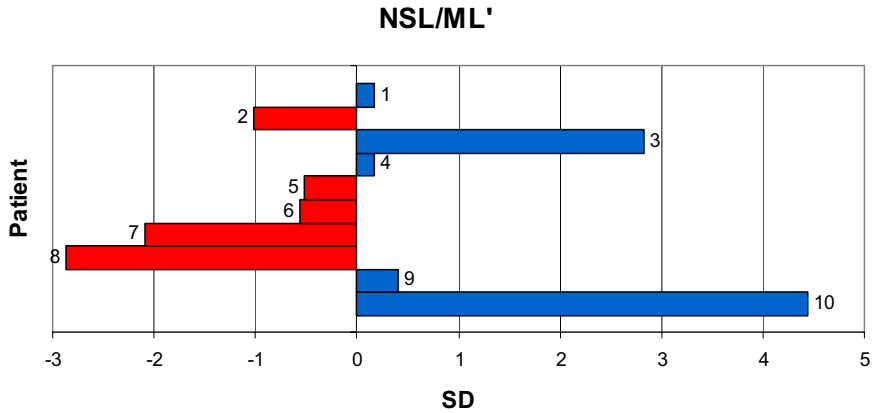

FIGURE 8. NSL/ML′ angle.

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angle was too large, and in the other half, it was too small (Figs 6-8).

When the ratio of anterior lower face height to anterior total face height was analyzed, it was inside the first SD in most of the patients (70%). Nevertheless, 2 patients showed ratios outside the second SD (Fig 9).

Concerning the y-axis (N-S-Gn), 30% of all patients showed angles outside the second SD, which indicates a trend in hyperdivergent growth patterns (Fig 10).

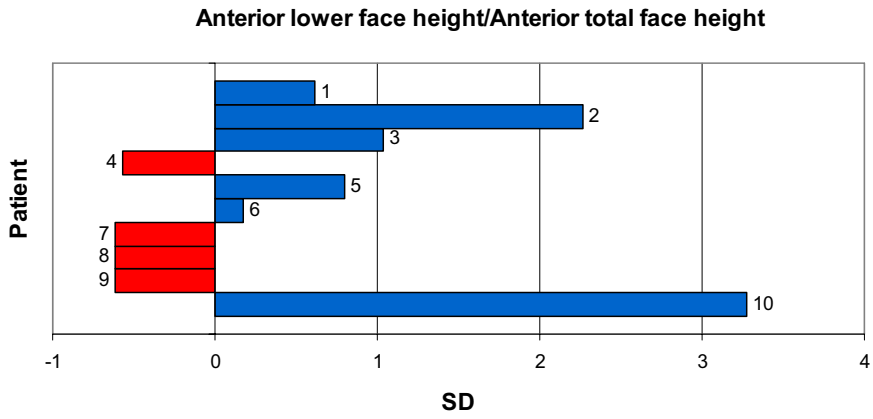

FIGURE 9. Ratio of anterior lower face height to anterior total face height.

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DENTAL PARAMETERS

A more homogeneous pattern was seen in the incisor inclination. In 80% of all patients, the upper central incisors are retroclined in relation to NL (and N-A) (Fig 11). The lower incisors are retroclined in relation to N-B in 70% of all patients and in relation to ML′ in 80% of all patients (Fig 12). Therefore the interincisal angle (IsL/IiL) is increased in 70% of all patients (Fig 13).

Discussion

Possibly because TGDCs are relatively rare congenital malformations of the thyroid apparatus and only some of these patients are operated on at an early age, a large patient size could not be obtained. For research purposes, it would have been preferable as well to have a more homogeneous patient group regarding age. In addition, a preponderance of male patients (n = 8) compared with female patients (n = 2) was apparent. The reason was probably the gender imbalance (28 male patients vs 12 female patients) already present in the surgical sample that was the basis for this study. However, female patients and male patients are equally affected by TGDCs.[9,15] Nevertheless, Greinwald et al[12] could show that female patients were more likely to be affected by TGDCs.

Several studies have shown that changes in mandibular position are related to hyoid bone changes and that hyoid bone position adapts to anteroposterior changes in the head posture.[4,16-19] The muscles of the tongue and the suprahyoid muscles have their insertions at the hyoid bone. From this point of view, it is possible that an interaction between growth direction and potential of the mandible, tongue position, and hyoid position exists. To evaluate this influence, either animal experiments where the hyoid bone is removed have to be conducted or patients need to be found in whom hyoid bone resection had to be carried out for different purposes before major craniomandibular growth takes place. Patients with TGDCs needing surgery represent a good possibility

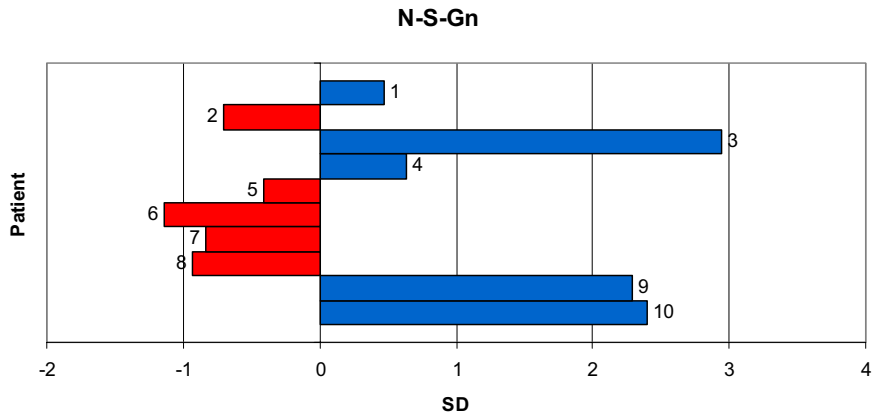

FIGURE 10. Y-axis.

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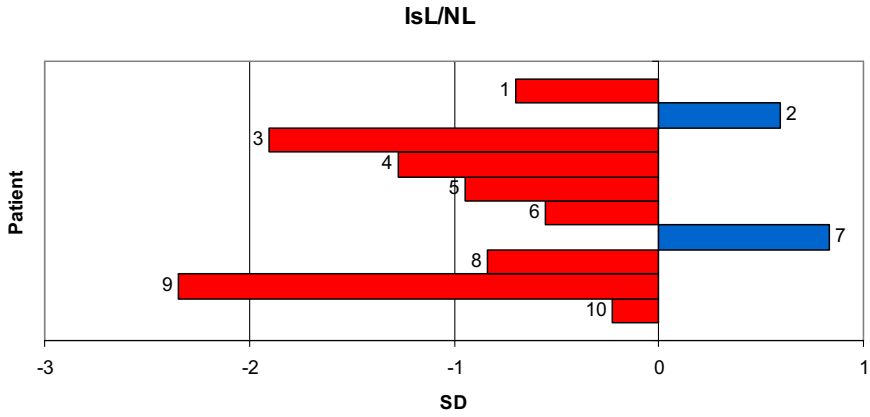

FIGURE 11. IiL/NL angle.

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in whom the influence of the partly resected hyoid bone (combined with muscular detachment and scarring of the throat region) on the hyoid-mandible-tongue equilibrium can be studied. On the other hand, a description of the disadvantages of the surgical method of Sistrunk[5] regarding craniomandibular growth has been lacking until now.

Cephalometric radiography and analysis comprise one of the most powerful tools in orthodontics to evaluate skeletal, dental, and soft tissue relationships of the craniofacial complex before starting orthodontic treatment or for research purposes. We decided to analyze the lateral cephalograms obtained from 10 patients with descriptive statistics and to compare the values with the growth standards of Bathia and Leighton[14] used as control parameters. Each patient was allocated according to his or her age when the lateral cephalogram was obtained and considering gender differences.

The sagittal parameters showed larger deviations for SNB than SNA from normal values described by Bathia and Leighton.[14] Patients with large deviations for SNB showed SNB angles that were too small. Hyoid resection more likely influences the mandible than the maxilla. Hyoid resection and/or scarring could produce a negative impact on mandibular growth either in a direct or an indirect manner through to dorso-caudal tongue retention.

However, comparisons of the ratio of mandibular length to maxillary length with the values of the Bathia and Leighton sample[14] showed that the mandible is too large compared with the maxilla in 80% of all patients. Therefore growth restriction of the mandible as a consequence of hyoid resection seems to be illogical in this patient sample. The possibility of mandibular growth deviation in the vertical plane could be an explanation for smaller SNB angles combined with increased mandibular length. Unfortunately, the vertical parameters tended to be heterogeneous in the way that some of the patients show a hyperdivergent pattern and others show a hypodivergent pattern.

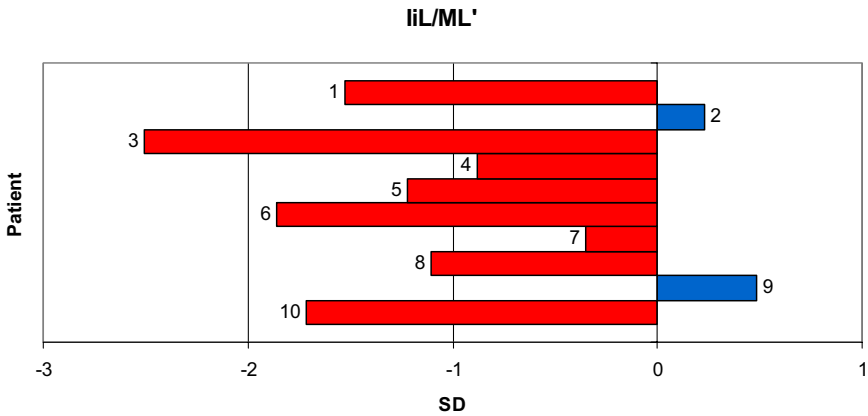

FIGURE 12. IiL/ML' angle.

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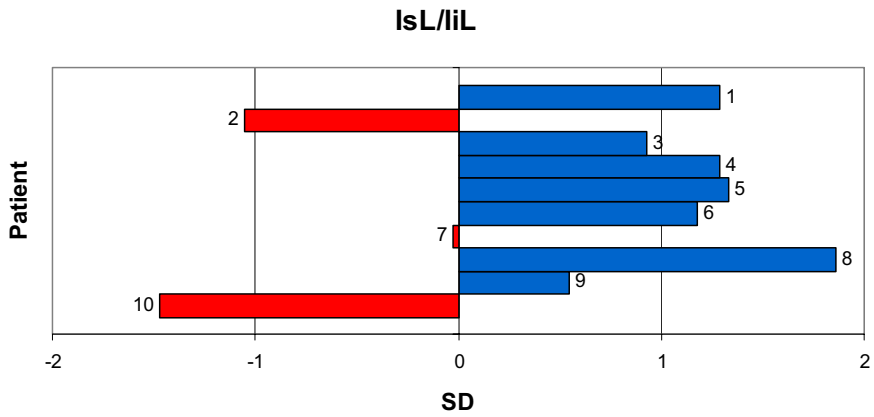

FIGURE 13. Interincisal angle.

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However, craniofacial growth is a very complex process influenced by genetics, habits, allergies, mouth breathing, orthopaedic appliances, and so on. The only environmental factor that could be excluded in this study was that none of the patients had orthodontic treatment before the lateral cephalogram was taken.

Concerning dental parameters, the majority of the 10 patients showed retroclined upper and lower incisors. A possible explanation could be that the tongue was kept posterior due to hyoid resection and scar tissue. Thereby the equilibrium between cheek or lip pressure to tongue pressure would have been changed. The decreased tongue pressure on the front teeth would then result in retroclined incisors.

To obtain further scientific evidence of the impact of hyoid resection on craniofacial growth, forthcoming studies should try to be more homogeneous in gender and age distribution, try to exclude environmental factors, and include larger patient populations with a prospective design.

The aim of this study was to evaluate retrospectively the influence of hyoid bone resection on craniofacial growth in early age according to Sistrunk[5] due to TGDCs. On the basis of the patients examined by use of cephalometric analysis, we have reached the following conclusions: Several vertical and horizontal skeletal and dental cephalometric parameters were shown to be different by trend when compared with control values. A possible negative impact on craniofacial growth potential and direction due to hyoid resection in early age according to Sistrunk[5] cannot be excluded.

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