Case Report

Surgical Orthodontic Correction of Acromegaly with Mandibular Prognathism

Takakaza Yagi, DDS, PhD[a]; Masayoshi Kawakami, DDS, PhD[b]; Kenji Takada, PhD, DDS[c]

Abstract: A male (30 years five months) who complained of mandibular prominence and masticatory dysfunction was diagnosed as a mandibular prognathic with acromegaly after cephalometric and endocrine examinations. The level of growth hormone (GH) subsequent to a transsphenoidal hypophysectomy had been controlled by medicines for about five years. Surgical orthodontic correction improved his occlusion and profile, but magnetic resonance imaging detected a recurrent adenoma in the cranial base during the retention period. The recurrence resulted in slight prognathic changes of the patient with a high level of GH. This is a case report of the treatment of an acromegalic patient discussing growth considerations that could influence the orthodontic treatment plan and long-term stability. (*Angle Orthod* 2004;74:125–131.)

Key Words: Acromegaly; Prognathism; Growth hormone; Retention; Adult phase growth

INTRODUCTION

Acromegaly is caused by an anterior pituitary tumor that secretes growth hormone (GH). Postpubertal overproduction of GH leads to highly disproportionate growth of the jaws and facial bones, which is mainly a result of periosteal bone apposition due to reactivation of the subcondylar growth zones.[1] When mandibular growth is the primary response and the dental occlusion begins to change, individuals with acromegaly often seek an orthodontic consultation. Enlargement of the ascending ramus and prominence of the mandible, chin, and lips are the most noticeable profile characteristics of acromegalic patients.[2,3] The excessive growth may stop when the tumor is removed or irradiated, but the skeletal deformity persists and often requires orthognathic surgery.[4] The treatment in adults should first include controlling the serum GH level by administering somatostatin followed by orthognathic surgery to correct the enlarged mandible if desired. However, the acromegaly patient has a substantial risk of recurrence of the pituitary adenoma, which causes growth changes in the facial skeleton even after transsphenoidal hypophysectomy.[5,6] We report the longitudinal records of the treatment of a patient with mandibular prognathism due to excess GH who had increased masticatory dysfunction and profile changes after surgical orthodontic treatment.

FIGURE 1. Initial hand-wrist photograph.

[a] Assistant Professor, Department of Orthodontics, Graduate School of Dentistry, Osaka University, Osaka, Japan.

[b] Assistant Professor, Department of Oral and Maxillofacial Surgery, Nara Medical University, Kashihara, Nara, Japan.

[c] Professor and Chair, Department of Orthodontics, Graduate School of Dentistry, Osaka University, Osaka, Japan.

Corresponding author: Takakaza Yagi, DDS, PhD, Department of Orthodontics and Dentofacial Orthopedics, Graduate School of Dentistry, Osaka University, 1-8 Yamadaoka Suita, Osaka 565-0871, Japan
(e-mail: tyagi@dent.osaka-u.ac.jp).

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FIGURE 2. Initial facial photographs.

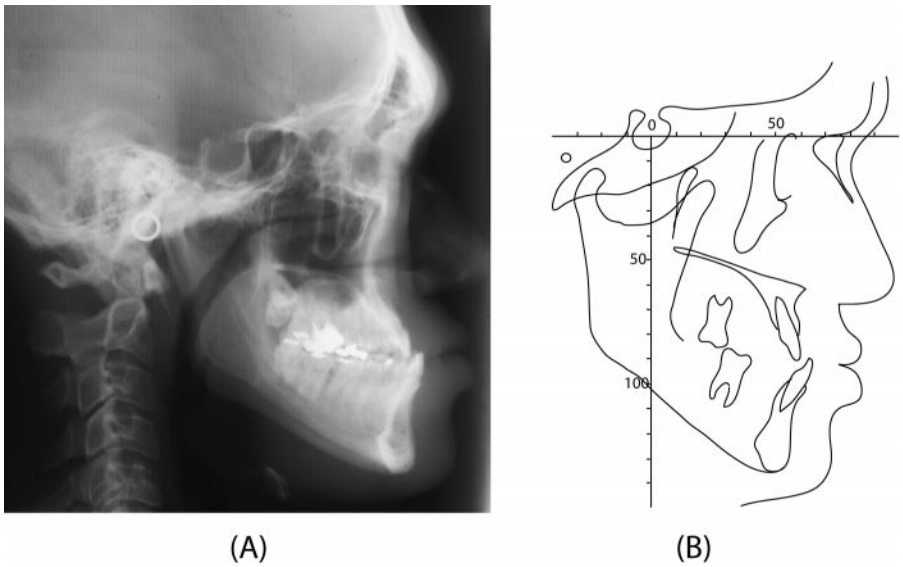

FIGURE 3. Initial (A) lateral cephalometric radiograph and (B) its tracing.

CASE REPORT

A male (30 years five months) was admitted to the Osaka University Dental Hospital with a chief complaint of mandibular protrusion and masticatory dysfunction. He had noticed enlargement of his hands and feet for the last 10 years (Figure 1). Five years earlier, a pituitary adenoma was detected, and the patient underwent microsurgical transsphenoidal hypophysectomy using the Hardy method.[7] The GH level improved from 14 to 2.97 ng/mL after completion of the surgical treatment. Two years later, however, the GH levels increased because of a recurrent adenoma that magnetic resonance imaging detected in the cranial base. Subsequently, his GH levels were controlled by bromocriptine mesilate (2.5 mg/day) and continuous dopamine agonists.

When the patient first visited the university hospital, he showed a convex facial appearance with prominent supraorbital ridges (Figure 2). The lateral profile indicated mandibular prognathism with enlarged lips and a bulbous nose. An enlarged sella turcica showed ballooning with a double floor and loss of definition of its bony outline. Cephalometric analysis revealed a Class III skel-

FIGURE 4. Initial intraoral photographs.

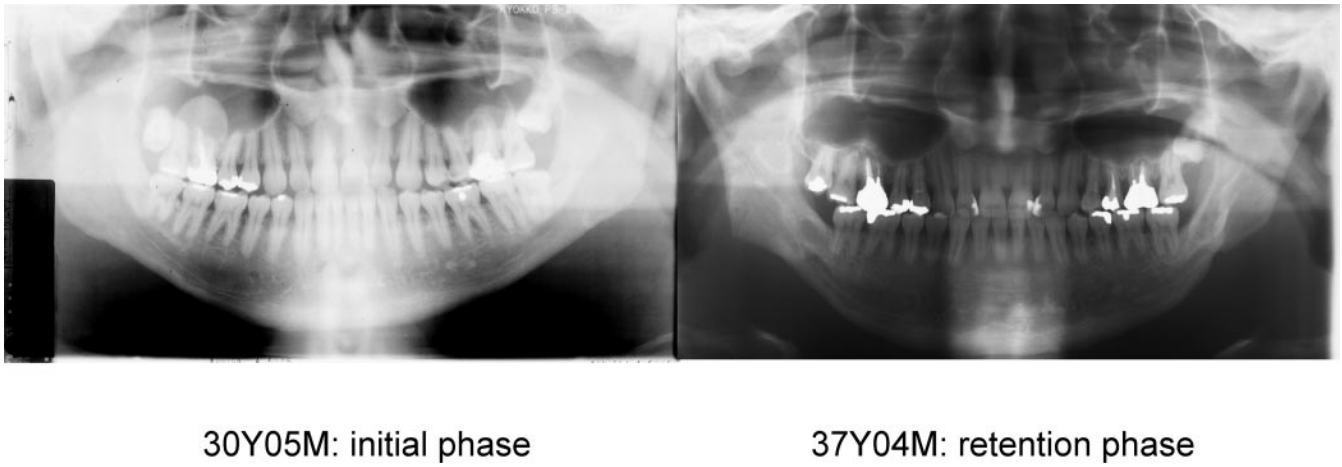

FIGURE 5. (A) Initial and (B) retention phase panoramic radiographs.

etal pattern (ANB = −4.2°) with an overjet of −4.7 mm. The molar relationship was Angle Class III with an arch length discrepancy of +14.6 mm. The radiograph and clinical examination did not reveal a macroglossia. There were no clinical symptoms of temporomandibular joint disorder, but the upper left first molar had a periapical lesion (Figures 3 through 5).

One year and seven months after the presurgical orthodontic treatment, including endodontic treatment of the upper molar and extraction of a lower third molar, a bilateral sagittal split ramus osteotomy (BSSRO) was performed to correct the negative overjet. A postsurgical increase in GH levels returned to a normal level by administrations of the bromocriptine mesilate. Eight months after surgery, all bands and brackets were removed, and removable type retainers were placed in the upper and lower arches (Figure 6).

Posttreatment results showed correction of the anterior crossbite and the mandibular excess. Facial esthetics was improved, and lip competence was obtained (Figure 7). Posttreatment orthodontic records revealed a well-interdigitated and aligned dentition, with a Class I occlusion on both sides. The profile was straight, but the overall appearance of the face remained characteristic of the appearance of acromegaly. His GH levels had gradually been increasing to over 5 μg/L during the retention phase. The additional administration of bromocriptine mesilate did not improve the level of GH.

Thus, a second transsphenoidal hypophysectomy was planned during the retention period, and the GH levels were

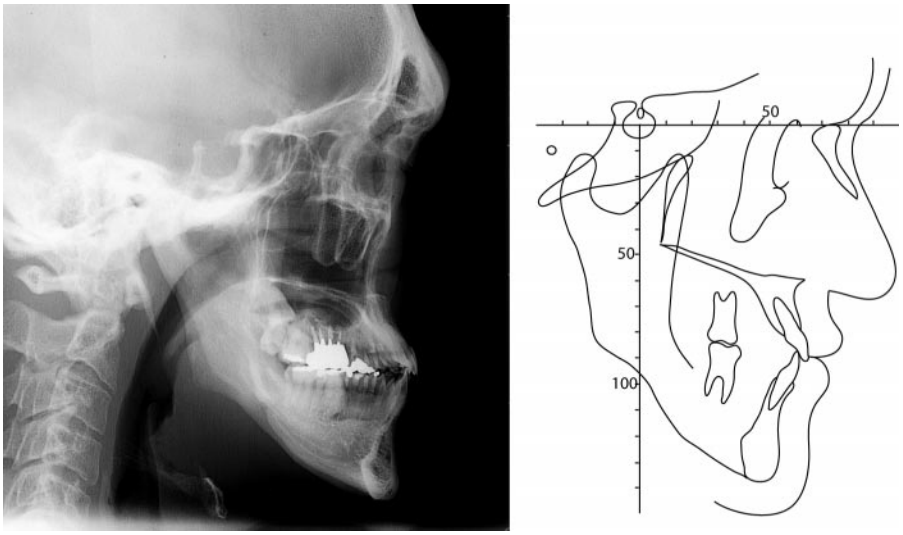

FIGURE 6. (A) Lateral cephalometric radiograph and (B) its tracing after active treatment.

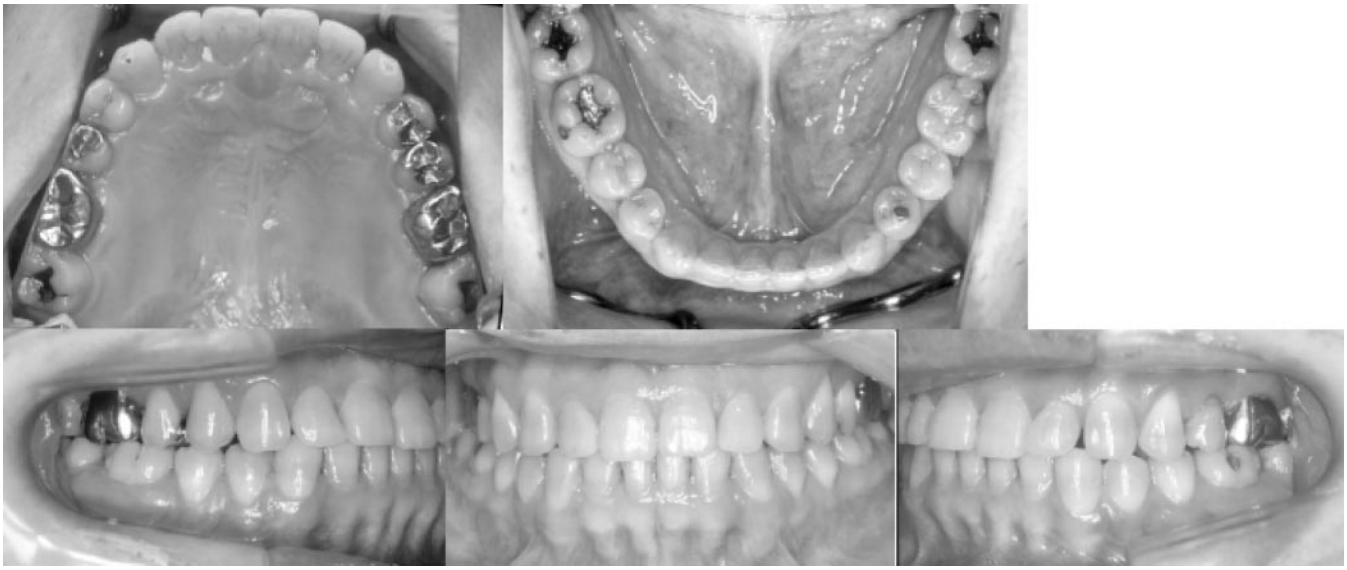

FIGURE 7. Intraoral photographs after active treatment.

maintained with octreotide acetate 200 μg/day to diminish GH secretion from the cells of the pituitary body. Three years later, the altered administration of octreotide acetate to the levothyroxine sodium (thyroid hormone) 50 μg/day resulted in a basal GH less than 5 μg/L. During these changes in GH levels, although the overjet and overbite were maintained (Figures 9 through 11), cephalometric comparison between postactive treatment and the retention demonstrated the increased SNB from 76.0° to 79.0° and decreased ANB from −3.0° to −6.5°. Superimposition of the cephalograms showed that his mandibular growth change remained, suggesting downward growth of the mandible with lingual inclination of the lower incisor (Figure 12; Table 1).

DISCUSSION

Changes in the face of acromegalic patients are often subject to skeletal and endocrine surgery.[8] Transsphenoidal selective hypophysectomy is the most efficient and widely used method to treat acromegaly.[9] It provides an initial favorable outcome for acromegalic patients, but the reported long-term cure rates have been variable. Serri et al[10] concluded that the overall cure rate was 68%. They stated that

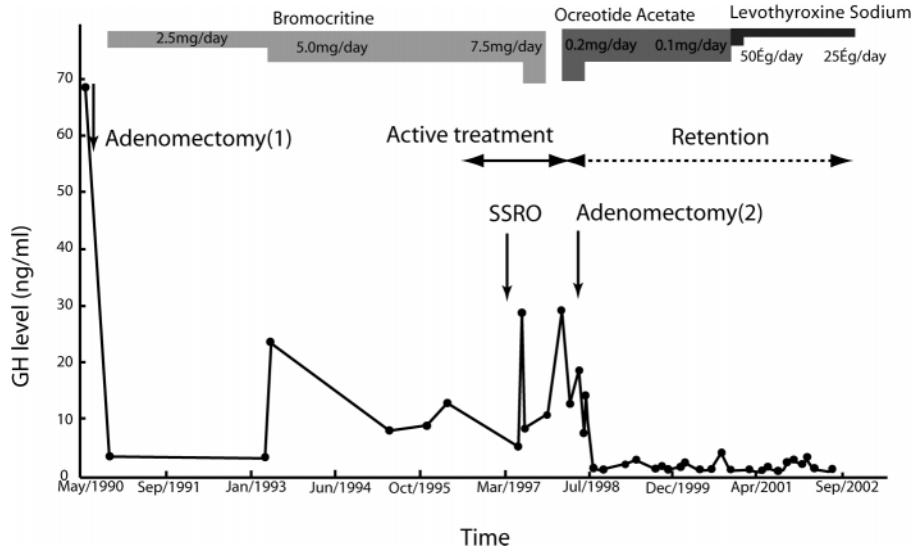

FIGURE 8. Changes in the GH level and clinical course of orthodontic and medical treatments.

FIGURE 9. Facial photographs at the retention phase.

acromegaly might recur after a successful surgery and adenoma removal, and it cannot be predicted by normal postoperative GH levels and dynamics.

As for craniofacial morphology, no significant changes were observed in 72% of the patients after removal of a pituitary adenoma. A nonspecific growth pattern of the facial skeleton could not be shown in 10 of 31 patients. However, other patients showed significant changes in the lower jaw. The ascending ramus in addition to the mandible body was elongated, and the main contribution to the elongation was made by chin prominence and condyle.[11] Thus, there is a great variability in expressivity of the signs and symptoms of acromegaly. Postpubertal patients have a potential to express a recurrence of GH levels and a prognathic facial pattern after removal of pituitary adenoma.

The present patient visited us for correction of his anterior crossbite with mandibular protrusion. He had already received a transsphenoidal hypophysectomy, and the level of GH had been controlled by administration of somatomedin C. Significant growth of the mandible, similar to previous studies, was shown at the beginning of preoperative orthodontic treatment, although the level of GH gradually increased. BSSRO surgery achieved facial and skeletal improvement, but because high levels of serum GH remained, a second hypophysectomy was performed. Thereafter, endocrinological and orthodontic examinations identified no relapse of the GH level or his occlusion with the craniofacial structure in the patient.

One study reported that the surgical procedures and the postoperative course of these patients were uneventful and

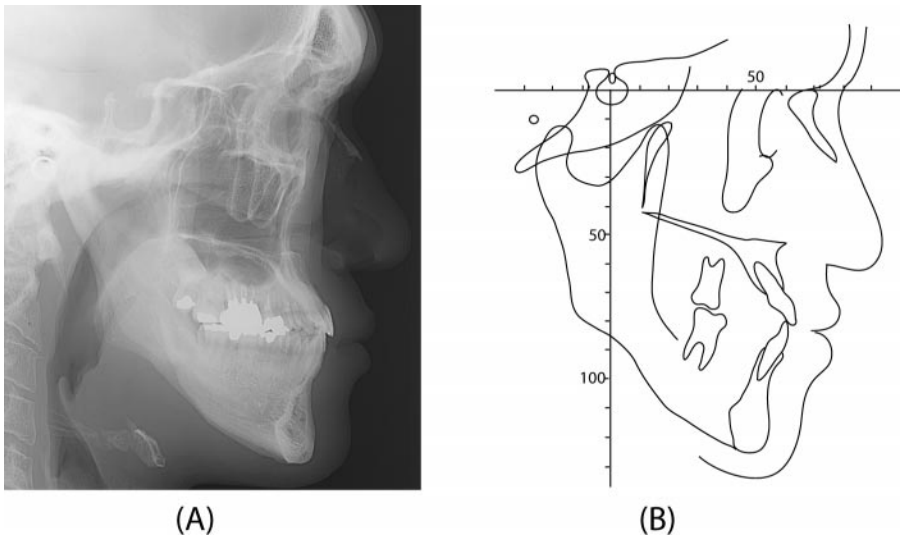

FIGURE 10. (A) Lateral cephalometric radiograph and (B) its tracing at the retention phase.

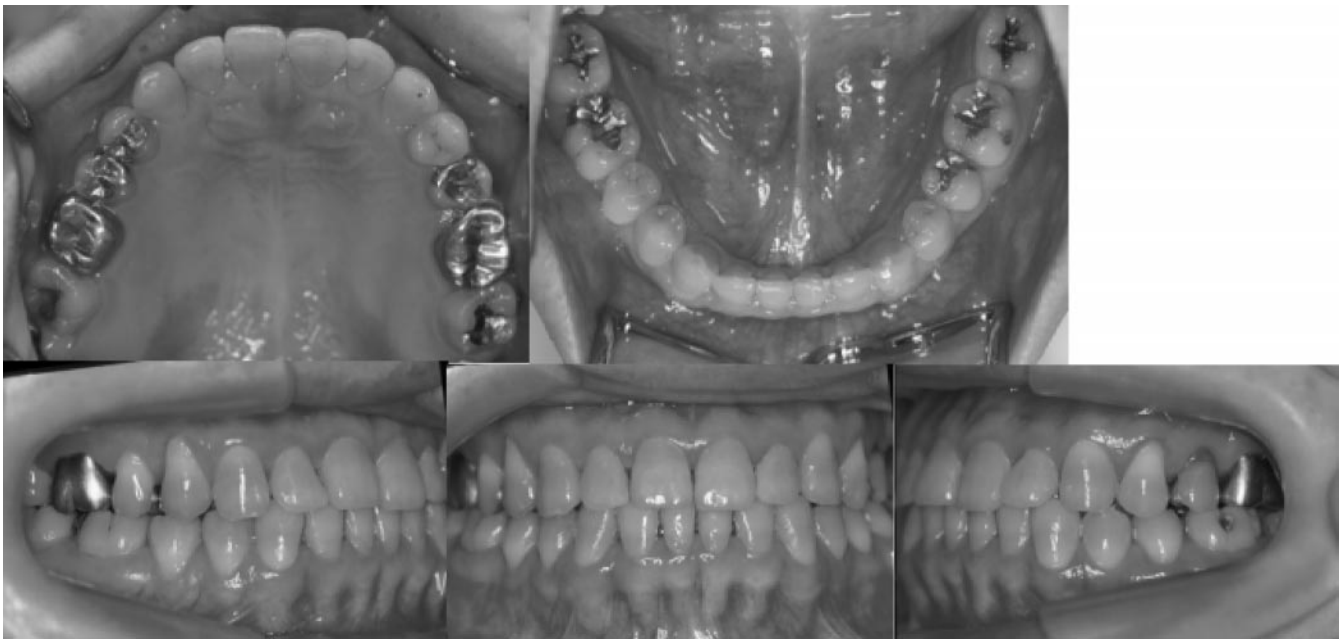

FIGURE 11. Intraoral photographs at the retention phase.

did not deviate from those of otherwise healthy patients.[3] It is accepted that after the reestablishment of endocrine balance, mandibular osteotomies should be considered to reestablish the premorbid facial contours and occlusion.[3] However, it is evident that an acromegalic patient has a substantial risk of recurrence of the GH level and growth changes of the facial skeleton after extirpation of the adenoma and hormonal control. In view of the present result, if there is any moderate reversibility after normalizing the GH level, it could hardly be seen clinically or appreciated by the patient.[12] Otherwise, a condylectomy may be presented as one of the surgical procedures to prevent unpredictable growth of the mandible, but this procedure may later lead to lost jaw function.[13] Because acromegaly is a persistent or recurrent disease, its orthodontic correction, attention to endocrine control, and facial skeletal growth changes may continue after puberty, and this should determine the timing of the treatment.

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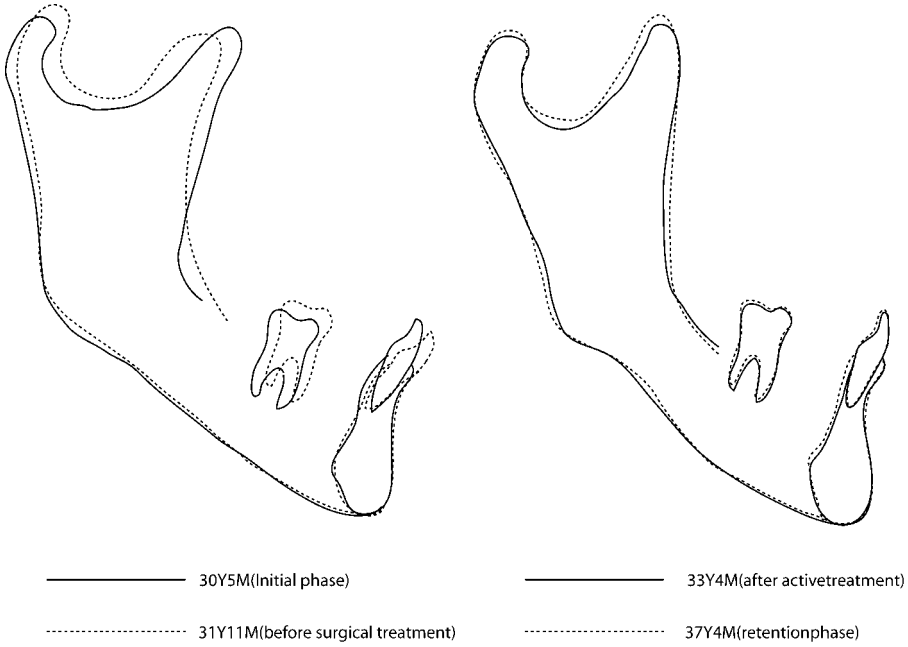

FIGURE 12. Superimpositions on the mandibular plane at Me from 30 years five months to 37 years four months (4th stage).

TABLE 1. Cephalometric Analysis

	Initial phase 30 y 5 mo	Before surgical 31 y 11 mo	After active 33 y 4 mo	Retention phase 37 y 4 mo
SNA (°)	72.5	72.5	73.0	73.5
SNB (°)	79.0	81.0	76.0	79.0
ANB (°)	−6.5	−8.5	−3.0	−5.5
Gonial angle (°)	131.5	132.0	145.0	146.0
S-N (mm)	78.0	78.0	78.0	78.0
Go-Me (mm)	88.0	89.0	79.0	80.0
Ar-Go (mm)	58.0	60.0	54.0	55.5
Ar-Me (mm)	135.0	138.0	127.0	128.0
Overjet (mm)	−7.0	−11.0	3.5	3.0
Overbite (mm)	1.5	−0.5	3.0	3.0

REFERENCES

1. Tornes K, Gilhuus-Moe O. Correction of jaw deformities subsequent to treatment of acromegaly. *Int J Oral Maxillofac Surg.* 1986;15:446–450.
2. Takakura M, Kuroda T. Morphologic analysis of dentofacial structure in patients with acromegaly. *Int J Adult Orthod Orthognath Surg.* 1998;13:277–288.
3. Clayton RN. New developments in the management of acromegaly. *J Endocrinol.* 1997;155(suppl 1):523–529.
4. Proffit WR. The development of dentofacial deformity. In: Proffit WR, White RP Jr, Sarver DM, eds. *Contemporary Treatment of Dentofacial Deformity.* Philadelphia, Pa: Mosby Yearbook; 2002: 51–53.
5. Hardy J. Transspenoidal hypophysectomy. *J Neurosurg.* 1971;34: 582–594.
6. Long H, Beauregard H, Somma M, et al. Surgical outcome after repeated transsphenoidal surgery in acromegaly. *J Neurosurg.* 1996;85:239–247.
7. Beauregard H, Somma M, et al. The treatment of acromegaly. *Union Med Can.* 1993;122:487–495.
8. Jackson IT, Meland NB, Keller EE, Sather AH. Surgical correction of the acromegalic face. A one stage procedure with a team approach. *J Craniomaxillofac Surg.* 1989;17:2–8.
9. Hardy J, Somma M. Acromegaly: Surgical treatment by transsphenoidal microsurgical removal of the pituitary adenoma. In: Tindall GT, Collins WS, eds. *Clinical Management of Pituitary Disorders.* New York, NY: Raven Press; 1979:209–211.
10. Serri O, Somma M, Comtois R, Rasio E, Beauregard H, Jilwan N, Hardy J. Acromegaly: biochemical assessment of cure after long term follow-up transsphenoidal selective adenomectomy. *J Clin Endocrinol Metab.* 1985;61:1185–1189.
11. Kunzler A, Farmand M. Typical changes in the viserocranium in acromegaly. *J Craniomaxillofac Surg.* 1991;19:332–340.
12. Landolt AM, Strebel P. Technique of transsphenoidal operation for pituitary adenomas. *Adv Tech Stand Neurosurg.* 1980;7: 119–177.
13. Ueki K, Nakagawa K, Matsumoto N, Yamamoto E. An experience of surgical reduction in a case with acromegaly. *Jpn J Jaw Deformities.* 1996;6:170–175.