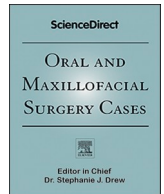




ELSEVIER

Contents lists available at [ScienceDirect](https://www.sciencedirect.com)

Oral and Maxillofacial Surgery Cases

journal homepage: www.oralandmaxillofacialsurgerycases.com

Surgical management of the acromegalic face: Could the aesthetic improvement of the face influence the patient's QoL? Combined surgical approach

Andrea Cassoni ^{a,b}, Resi Pucci ^{a,*}, Paolo Priore ^a, Maria Teresa Fadda ^{a,b},
Valentino Valentini ^{a,b}

^a Dipartimento di Scienze Odontostomatologiche e Maxillofacciali, Sapienza Università di Roma, Via Caserta, 6, 00161, Rome, Italy

^b U.O.D. Chirurgia Oncologica Ricostruttiva Maxillo Facciale; D.A.I. Testa e Collo, Azienda Ospedaliero-Universitaria Policlinico Umberto I di Roma, Viale del Policlinico 155, 00161, Rome, Italy

ARTICLE INFO

Keywords:

Acromegaly
Orthognathic surgery
Partial glossectomy
Rhinoplasty

ABSTRACT

Acromegaly is a chronic and progressive disease related to a disorder of growth hormone production. It may present with a variety of clinical signs and a dento-facial deformity and this results in a loss of self-esteem and a reduction of Quality of Life.

Presentation of case: A 38-years-old male patient, affected by acromegaly with class III malocclusion, noticeable nose deformity and macroglossia was treated. Bi-maxillary orthognathic surgery and partial glossectomy have been performed in one-step surgery. Open rhinoplasty was done in a second step. Both dento-skeletal class III and restoration of the facial appearance have been solved. Dento-skeletal class III was completely solved together with the restoration of the facial appearance and the patient satisfaction has been achieved.

Conclusions: This case report describes the successful and stable treatment of an adult patient affected by acromegaly.

1. Introduction

Pathogenesis of acromegaly is well known as is the effect that GH and IGF have on tissues and organs. It is known that the face is not spared by the effects of this chronic and progressive pathology. Furthermore, even when the cause is identified and treated the effects that these hormones have had on the skeleton, skin, cartilage and muscles of the face are irreversible [1]. Acromegaly has a subclinical onset so often at diagnosis patients have even serious complications such as: diabetes mellitus, hypertension and respiratory/cardiac failure [2,3]. However acromegaly is frequently associated with altered facial appearance at the time of diagnosis and sometimes new diagnoses of acromegaly are detected by others specialists than endocrinologists. Acromegalic patients have typical face, nose, ears and jaw protruded, with a slightly open mouth due to labial incompetence [3,4]. Macroglossia frequently is accompanied by obstructive sleep apnea syndrome (OSAS) and also influences the orthognathic treatment, because that may cause a postoperative relapse [3,4]. These disturbances can be associated with impaired self-esteem and body image distortion. These disorders of course affect the quality of life of the patient [6,7]. A multidisciplinary approach is mandatory not only in the treatment of endocrinological

* Corresponding author. Sapienza Università di Roma, Dipartimento di Scienze Odontostomatologiche e Maxillofacciali, Via Caserta 6, 00161, Roma.

E-mail addresses: resi.pucci@gmail.com, resi.pucci@uniroma1.it (R. Pucci).

<https://doi.org/10.1016/j.omsc.2020.100150>

Received 8 April 2020; Accepted 14 April 2020

Available online 25 April 2020

2214-5419/© 2020 Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

disorder but especially in the management of patient's sequelae.

1.1. Presentation of Case

A 38-years-old male patient suffering from acromegaly was admitted in our Department in 2016 for evaluation and treatment. He previously had been diagnosed with GH-secreting pituitary adenoma. Tumor had been resected in 2008 and he didn't need medical therapy. Patient presented the typical acromegalic face (Fig. 1). A lateral cephalogram showed a skeletal class III relationship SNA 85.41°; SNB 96.0°; ANB -10.59° (Fig. 2A) He displayed an anterior open bite, involving a -3.79 mm overbite and a -14.31 mm overjet. Polysomnography (PSG) revealed a moderate OSAS: Apnea-Hypopnea Index (AHI), 28.2/h; apnea index (AI) 27.5/h, hypopnea index (HI) 0.6/h. and Oxygen Desaturation Index (3% ODI) 17,84/h. Cone Beam Computer Tomography (CBCT) was acquired and 3D Virtual Surgical Plan was planned. Surface scanning of each dental model arch was achieved, thereby producing an STL file that were fused with the 3D reconstruction. 3D Virtual Surgical Plan was performed and osteotomies and movement of maxilla and mandible have been simulated by the software. Bi-maxillary orthognathic surgery was performed, including Le Fort I osteotomy advancement and bilateral sagittal splint ramus osteotomy. During the first surgical step, partial glossectomy was performed using the tongue reduction Harada-Enomoto method [8]. The open rhinoplasty was performed in 2018 included the tip-plasty, septoplasty and nasal bone osteotomies to correct the abnormal growth and shape of the nose [9].

Dento-skeletal class III was solved together with the restoration of the facial appearance (Figs. 2B and 3). After orthognathic treatment SNA, SNB, ANB were 89.93°; 90.20°; -0.27° respectively (Fig. 2). A CBCT scan was performed two years after surgery and confirmed the stable results obtained. The PSG exam, repeated 1-year after surgery showed a mild OSAS and an improvement of all parameters AHI, AI, HI were 11.4, 9.8 and 1.5.

2. Discussion

The mandible is the most affected part of the face and the length is correlate with disease duration [1-3]. Dentists or orthodontists should especially consider acromegaly when symmetrical bilateral mandibular prognathism is noticed. In particular, acromegaly should be considered when there is a need for new dental prostheses and breaking of dental crowns at the same time [3]. There are cephalometric points that present typical anomalies in patients with acromegaly [5]. In the patient SNB and ANB-angle were 96.0° and 89.93° and ANB angle was -10.59° before orthognathic treatment. The WHO defines Quality of Life as the individual's perception of his/her position in life in the context of the culture and value systems in which he/she lives, and in relation to his/her goals, expectations, standards and concerns [7]. The self-perception have an important impact on quality of life. The combined surgical approach allows to restore aesthetic and functional problems in this adult patient. The goals of the whole treatment for the patient were



Fig. 1. Pre-operative patient facial appearance. The frontal, lateral and spontanous smile view. Pre-operative anterolateral, frontal and lateral intraoral occlusal view. Published with the patient's consent.

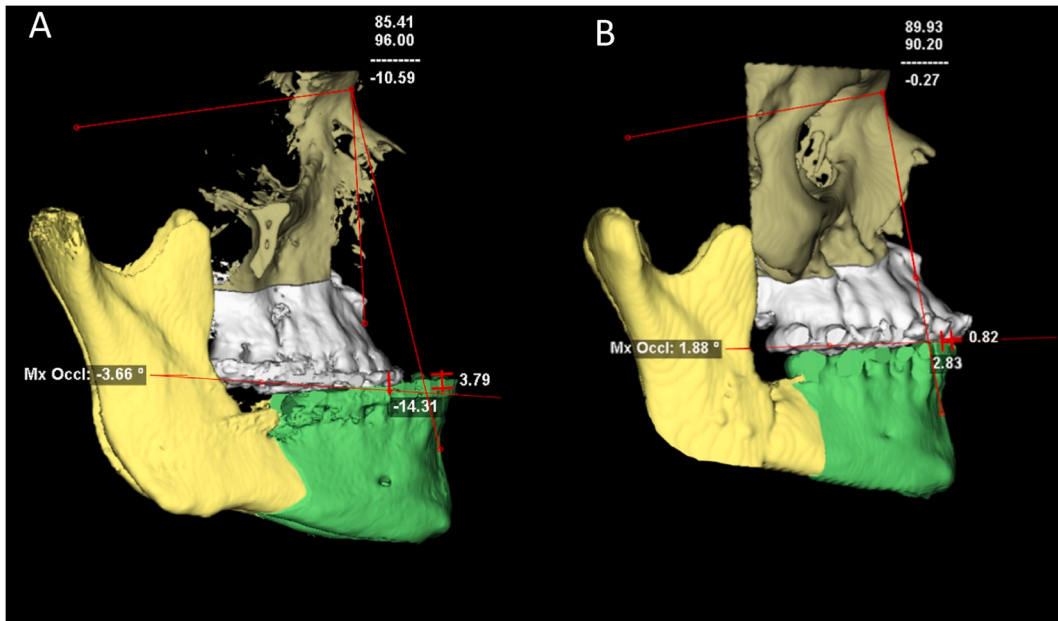


Fig. 2. Pre-operative and post-operative CBCT 3D lateral view. **A:** Pre-operative CBCT 3D reconstruction lateral view, that show cephalometric measures: SNA 85.41°, SNB 96.00°, ANB-angle -10.59°, overbite -3.79 mm and overjet -14.31 mm. **B:** Post-operative CBCT 3D reconstruction lateral view, that show cephalometric measures: SNA 89.93°, SNB 90.20°, ANB-angle -0.27°, overbite -0.82 mm and overjet -2.83 mm.



Fig. 3. Post-operative patient facial appearance. The frontal, lateral and spontaneous smile view. Post-operative anterolateral, frontal and lateral intraoral occlusal view. Published with the patient's consent.

to correct the skeletal malocclusion, conserving the motor and sensory functions of the tongue, reduce the respiratory problem and finally to improve the soft tissue profile. The decision to make contemporary the orthognathic treatment and partial central glossectomy is related to prevent relapse caused by an enlarged or dysfunctional tongue. Several techniques of tongue reduction have been reported however we prefer to preserve the tip is the most mobile and sensitive portion of the tongue [10]. The partial glossectomy operation was performed by Harada-Enomoto method [8] whereby the center of the tongue and part of the base are removed, bringing an overall shortening of the tongue vertically and transversely, sparing the tip intact, to prevent the paralytic symptoms that hit the tip of the tongue, keeping the sense of taste intact. A CBCT scan was performed two years after surgery and confirmed the stable results obtained, the images show an improvement in the shape of the upper airways. This was confirmed by a PSG exam, repeated one year after surgery, that showed a mild OSAS. After the orthognathic treatment, harmonizing the bone segments, the shape and size of the patient's nose has become even more evident. Therefore open rhinoplasty was necessary to reduce the size and shape of the nose, this surgical procedure is perhaps the most complex cosmetic procedure performed [9].

3. Conclusions

Acromegaly is associated with typical altered facial appearance and cosmetic deformities are only partially reversible upon disease control that can lead to a lack of self-confidence and psychological trauma while decreasing quality of life as already reported in the literature. The clinical outcomes (Fig. 2D and Fig. 3) of this patient support the decision to treat him with combined surgical approach and aesthetic refinement in a second time. Often treating the illness of these patients does not mean improving their quality of life. Therefore the therapeutic process of these subjects should be integrated with the evaluation of the maxillofacial surgeon.

Author contribution

Andrea Cassoni: Conception and design of study, Critical revision, Final approval. Resi Pucci: Writing- Original draft preparation, Literature search. Paolo Priore: Acquisition of data, Investigation, Literature search. Maria Teresa Fadda: Supervision, Critical revision. Valentino Valentini: Reviewing, Editing, Final approval.

Consent

Written informed consent for publication of the clinical details and clinical images was obtained from the patient.

Funding

None.

Declaration of competing interest

None.

References

- [1] Katznelson L, Laws Jr ER, Melmed S, Molitch ME, Murad MH, Utz A, Wass JA, Endocrine Society. Acromegaly: an endocrine society clinical practice guideline. *J Clin Endocrinol Metab* 2014 Nov;99(11):3933–51. <https://doi.org/10.1210/jc.2014-2700>. Epub 2014 Oct 30. PubMed PMID: 25356808.
- [2] García-Álvarez M, Climent V. Sleep apnea and cardiovascular complications of the acromegaly. Response to the medical treatment. *Minerva Endocrinol* 2019 Jun;44(2):159–68. <https://doi.org/10.23736/S0391-1977.18.02930-9>. Epub 2018 Nov 26. Review. PubMed PMID: 30482010.
- [3] Herrmann BL, Mortsch F, Berg C, Weischer T, Mohr C, Mann K. Acromegaly: a cross-sectional analysis of the oral and maxillofacial pathologies. *Exp Clin Endocrinol Diabetes* 2011;119(1):9–14.
- [4] Nakano H, Mishima K, Matsushita A, Suga H, Miyawaki Y, Mano T, Mori Y, Ueyama Y. Orthognathic surgery in an acromegalic patient with obstructive sleep apnea syndrome. *Sleep Breath* 2013 Mar;17(1):29–32. <https://doi.org/10.1007/s11325-012-0711-5>. Epub 2012 May 2. PMID: 22549813.
- [5] Bavbek NC, Tuncer BB, Tuncer C, Gungor K, Ozkan C, Arslan E, Altinova AE, Akturk M, Toruner FB. Cephalometric assessment of soft tissue morphology of patients with acromegaly. *Aust Orthod J* 2016 May;32(1):48–54. PMID: 27468591.
- [6] Paisley AN, Rowles SV, Roberts ME, Webb SM, Badia X, Prieto L, Shalet SM, Trainer PJ. Treatment of acromegaly improves quality of life, measured by AcroQoL. *Clin Endocrinol* 2007 Sep;67(3):358–62. Epub 2007 Jun 6. PubMed PMID: 17555502.
- [7] Rowles SV, Prieto L, Badia X, Shalet SM, Webb SM, Trainer PJ. Quality of life (QOL) in patients with acromegaly is severely impaired: use of a novel measure of QOL: acromegaly quality of life questionnaire. *J Clin Endocrinol Metab* 2005;90:3337–41.
- [8] Balaji SM. Reduction glossectomy for large tongues. *Ann Maxillofac Surg* 2013 Jul;3(2):167–72. <https://doi.org/10.4103/2231-0746.119230>. Review. PubMed PMID: 24205477; PubMed Central PMCID: PMC3814666.
- [9] Gassner HG. Structural grafts and suture techniques in functional and aesthetic rhinoplasty. *GMS Curr Top Otorhinolaryngol, Head Neck Surg* 2010;9:Doc01. <https://doi.org/10.3205/cto000065>. Epub 2011 Apr 27. PubMed PMID: 22073105; PubMed Central PMCID: PMC3199824.
- [10] Matsumoto K, Morita K, Jinno S, Omura K. Sensory changes after tongue reduction for macroglossia. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2014 Jan;117(1):e1–2. <https://doi.org/10.1016/j.oooo.2012.02.037>. Epub 2012 Aug 15. PubMed PMID: 22901656.