

Fig. 2. Segmentations of the patient's skull derived from the direct and 18-month postoperative CT scans were aligned using the Best Fit Alignment tool in Geomagic Studio 2012 software (3D Systems, Rock Hill, SC, USA) whilst moving along an identical CAD file of the implant. Note the nearly perfect alignment of direct postoperative (grey) and 18-month follow-up (blue) implant position. This nearly perfect alignment indicates good clinical implant stability.

signs of inflammation and diagnose these early. Our patient has reported no discomfort or pain. No signs of the development of peri-implant mucositis or peri-implantitis have been observed by the dentist or oral hygienists during the 18 months of follow-up to date since placement of the psSPI.

The additively manufactured sub-periosteal jaw implant – AMSJI – developed by Mommaerts is a valid treatment option for excessive maxillary bone loss, poor bone quality, and maxillary pneumatization<sup>3</sup>. However for oncology cases, such as our case, an unmodified Mommaerts approach is not applicable, as that approach relies on vertical support and fixture on maxillary and palatal bone structures that are often no longer present.

#### Funding

None.

#### Competing interests

None declared.

#### Ethical approval

Not required.

#### Patient consent

Written consent was obtained.

N. Vosselman\*  
B.J. Merema  
G.M. Raghoobar  
A. Vissink

*Oral and Maxillofacial Surgery and Maxillofacial Prosthetics, University of Groningen and University Medical Centre Groningen, Groningen, The Netherlands*

\*Address: N. Vosselman, Oral and Maxillofacial Surgery and Maxillofacial Prosthetics, University of Groningen and University Medical Centre Groningen, PO Box 30.001 - BB70, 9700 RB Groningen, The Netherlands. E-mail: n.vosselman@umcg.nl

E-mail address: n.vosselman@umcg.nl (N. Vosselman)

#### References

1. Vosselman N, Merema BJ, Schepman KP, Raghoobar GM. Patient-specific sub-periosteal zygoma implant for prosthetic rehabilitation of large maxillary defects after oncological resection. *Int J Oral Maxillofac Surg* 2019;48:115–7.
2. Gibbs CH, Lundeen HC. Jaw movements and forces during chewing and swallowing and their clinical significance. In: Lundeen HC, Gibbs CH, editors. *Advances in occlusion*. Boston: John Wright PSG Inc.; 1982. p. 2–32.
3. Mommaerts MY. Evolutionary steps in the design and biofunctionalization of the additively manufactured sub-periosteal jaw implant 'AMSJI' for the maxilla. *Int J Oral Maxillofac Surg* 2019;48:108–14.

#### Beckwith–Wiedemann syndrome

I read with great interest the paper titled, “Tongue reduction in Beckwith–

Wiedemann syndrome: outcome and treatment algorithm” by Naujokat et al., published in this journal in January 2019<sup>1</sup>. I would like to bring to the authors' attention our article published in 1995, where we looked at dentofacial growth following early partial glossectomies in patients with Beckwith–Wiedemann syndrome<sup>2</sup>. I concur with the authors that early tongue reduction in cases with significant macroglossia is beneficial. In our study we clearly showed that the abnormal craniofacial morphology is most often seen as an open bite with mandibular prognathism and inferior rotation. The maxilla was also involved in all cases, being shortened and rotated superiorly. The dentofacial morphology reverted to a more normal condition following tongue surgery (Figs 1 and 2). In addition, the abnormal growth is mainly caused by the macroglossia, as evidenced by the normalizing post-surgical changes. In addition, the mandibular condyle is a passive growth center stimulated by function and not an active growth center, as seen in long bone cartilaginous epiphyses. I applaud the authors on their conclusions regarding tongue function and hope that these additional cephalometric data will be beneficial.

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None.

#### Competing interests

None.

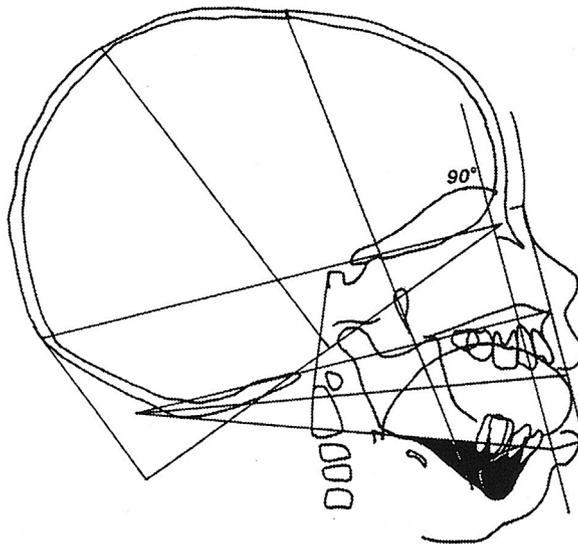


Fig. 1. Cephalometric study of a 2-year-old with Beckwith–Wiedemann syndrome and severe macroglossia. The mandible has been rotated down and back. In addition, the maxilla has been rotated superiorly and shortened vertically.

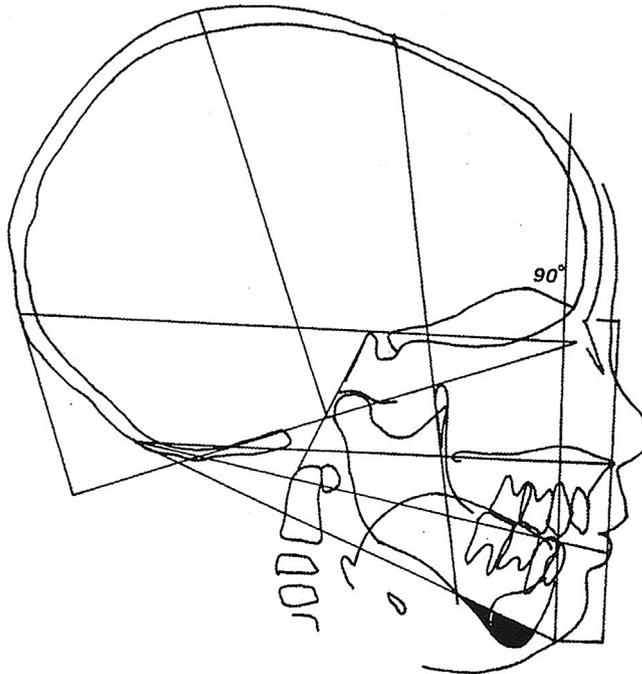


Fig. 2. Cephalometric study of the same child as in Fig. 1 at 5 years of age, following a partial glossectomy at 2 and 1/2 years of age. The mandible has rotated superiorly and the maxilla has elongated vertically, resulting in a more normal dentofacial skeleton.

*Ethical approval*

Not required.

*Patient consent*

Not required.

\*Address: Stephen A. Schendel, Stanford University, 1900 University Ave., Suite 101, E. Palo Alto, CA, 94303, USA. Tel.: +1 650 494 1000.

E-mail address: [sschendel@stanford.edu](mailto:sschendel@stanford.edu) (S.A. Schendel)

**References**

1. Naujokat H, Möller B, Terheyden H, Birkenfeld F, Caliebe D, Krause MF, Fischer-Brandies H, Wiltfang J. Tongue reduction in Beckwith–

Wiedemann syndrome: outcome and treatment algorithm. *Int J Oral Maxillofac Surg* 2019;48:9–16. <http://dx.doi.org/10.1016/j.ijom.2018.07.008>. Epub 2018 Jul 26.

2. Menard R, Delaire J, Schendel S. Treatment of the craniofacial complications of Beckwith–Wiedemann syndrome. *Plast Reconstr Surg* 1995;96:27–33.

**Response to the Letter to the Editor in response to the publication “Tongue reduction in Beckwith–Wiedemann syndrome: outcome and treatment algorithm”**

We are very grateful that our publication “Tongue reduction in Beckwith–Wiedemann syndrome: outcome and treatment algorithm”, published in January 2019 (Epub July 2018)<sup>1</sup>, has aroused attention and interest.

Having received the ‘Letter to the Editor’, we appreciate their endorsement of our article. We are also grateful for the supplementary cephalometric data from the 1995 publication. Based on the cephalometric data, the authors conclude that the abnormal growth is mainly caused by the macroglossia, as proven by the normalizing post-surgical changes. We agree, as our data also show that surgical tongue reduction exerts a positive influence on skeletal, dentoalveolar, and functional development, with a satisfactory long-term outcome in the majority of patients. Macroglossia is thought to be one of the main factors. However, taking all data into account, one must conclude that cephalometric development seems to be more complex and not solely influenced by the hyperplastic tongue. In our cohort, one of the largest examined so far, 42% of the patients received orthodontic treatment over the course of time and 8% required combined orthodontic–orthognathic interventions, despite a previous sufficient reduction of the tongue.

We hope that the published treatment algorithm, as well as the supplementary cephalometric data in this correspondence, will encourage many more practitioners to focus on improving the treatment of Beckwith–Wiedemann syndrome in order to achieve optimal results and positive dentofacial development.

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*Competing interests*

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S.A. Schendel\*  
Stanford University, Palo Alto, California,  
USA