Long-term surgical-orthodontic management of hemimandibular hyperplasia

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Background: Hemimandibular hyperplasia (HH), also known as hemimandibular hypertrophy, is characterised by excessive unilateral three-dimensional growth of the mandible after birth. Vertical unilateral elongation of the mandible becomes clinically evident as a rare form of vertical facial asymmetry. Aberrant growth of the facial skeleton affects the developing dentition and the dental compensatory mechanism is usually unable to maintain optimal occlusal relationships. The resulting malocclusion is effectively managed by combined surgical-orthodontic care to address the facial, skeletal and dental problems that confront clinicians. Orthodontists are advised to assess patients with HH during the post-treatment retention stage for continuing mandibular growth and assess the stability of treatment outcomes with long-term follow-up and records as required.

Aim: To present a case of hemimandibular hyperplasia treated successfully by combined surgical-orthodontic care and evaluated for stability over a seven-year follow-up period.

Methods: Surgical-orthodontic management was accomplished in four stages: 1) pre-surgical orthodontic; 2) surgical; 3) post-surgical orthodontic; and 4) post-treatment orthodontic retention. Complete orthodontic records, including extra- and intra-oral photographs, study models, and cephalograms plus panoramic radiographs were taken at the pretreatment, post-treatment, and seven-year orthodontic retention time-points.

Results: Facial, skeletal and dental goals were achieved in the three planes of space and the long-term stability of the treatment results was shown during a post-treatment orthodontic retention period of seven years.

Conclusion: Hemimandibular hyperplasia is a true growth anomaly which may be managed effectively. Clinicians may expect successful long-term correction and stability by utilising a comprehensive surgical-orthodontic treatment approach.

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Introduction

Hemimandibular hyperplasia (HH), also known as hemimandibular hypertrophy, is a rare condition characterised by excessive unilateral, three-dimensional growth of the mandible after birth.\(^1\)\(^,\)\(^2\) The entire hemimandible (condyle, ramus, and mandibular body) is volumetrically enlarged on one side, terminating at the mandibular midline symphysis. Depending on the severity of HH, individuals are affected by varying degrees of vertical facial asymmetry, vertical skeletal asymmetry, and dental malocclusion. Severe forms of facial asymmetry are rare and, in total, represent approximately one-third of the number of patients seeking treatment for the correction of a dentofacial deformity.\(^3\)

In severe cases of HH, the rate, timing and duration of excessive mandibular growth is sufficient to produce a frontal cant to the occlusal plane, which is indicative of a vertical facial asymmetry.\(^3\)\(^,\)\(^4\) Excessive mandibular growth due to HH usually commences before adolescence and continues into adulthood. The vertical growth of the maxilla is also increased on the affected side, to the extent that the dental compensations for the developing vertical skeletal asymmetry are unable to maintain occlusal contact and a lateral open-bite develops on the affected side. Pressure from the growing hemimandible ‘squeezes’ the mandibular teeth into occlusion on the non-affected side, producing dental arches with a twisted corkscrew-like appearance.\(^1\) The posterior occlusion
on the non-affected side has a scissor-bite tendency as the mandibular teeth become tilted lingually during compensatory dentoalveolar growth and development. Surgical-orthodontic treatment provides a means of correcting the facial, skeletal, and dental problems of dentofacial deformity that are characteristic of HH, and a case is presented which has been treated successfully with orthodontics, bimaxillary surgery and evaluated for long-term stability via orthodontic records over a seven-year follow-up period.

**Diagnosis and aetiology**

A young Caucasian woman presented to the University of Western Australia Orthodontic Clinic because her ‘jaw growth was asymmetrical’ and her ‘face was lopsided’. The patient was aged 19 years, 7 months and the medical history was unremarkable. There was no family history of excessive asymmetric skeletal or facial growth. The family somatotype was ectomorphic; however, a brachyfacial craniofacial pattern was evident. The patient was studying at university and had previously undergone a condylar surgical procedure during adolescence in an attempt to limit the effect of condylar hyperplasia on mandibular growth. There was no history or symptoms of temporomandibular disorder (TMD) and the range of mandibular movements was unrestricted.

The initial pretreatment presentation of this case showed that the facial appearance was typical of HH with vertical elongation of the affected right side and counter-clockwise rotation when viewed from the frontal perspective. The lip commissure was sloping down towards the affected side (Figure 1a). Upon smiling, the gingival display on the right side was increased due to the underlying vertical elongation of the maxilla (Figure 1a). A frontal cant to the occlusal plane was evident as the patient occluded onto a tongue depressor at the pre-surgical record stage (Figure 2b). Skeletally, the cephalometric analysis indicated a hypodivergent Class I skeletal pattern with a mild retrognathic maxilla and mandible (Table I). Vertical skeletal asymmetry of the right maxilla and mandible was evident in the cephalogram (Figure 1c) and CT images (Figure 2a). Dentally, the patient presented with an Angle Class I-type malocclusion with increased overbite, a palatally ectopic right maxillary canine and severe upper arch irregularity (Figure 1b). A right lateral open-bite and a left scissor-bite tendency were evident in the canine/premolar regions. The upper and lower dental arch forms were asymmetrical and presented with a corkscrew-like appearance. A maxillary Bolton deficiency of 3 mm was measured primarily due to the peg-shaped upper right lateral incisor.

A full orthodontic examination, photographic records and radiographic examination indicated that a positive diagnosis of HH could be made. Serial lateral cephalometric superimpositions indicated that facial growth had ceased, and 99mTc bone scanning

| Table I. Cephalometric analysis. |
|----------------------------------|
| **Standard** | **Pre-tx** | **Post-tx** | **7-year retention** |
| Cranial base | SN-H (%) | 10 | 4 | 5 | 5 |
| Maxilla | SNA (%) | 81 | 78 | 78 | 78 |
| Mandible | SNB (%) | 78 | 77 | 80 | 80 |
| Basal arch relations | ANB (%) | 2.5 | 1 | -2 | -2 |
| | Vwits (mm) | 0 | -2 | -3 | -3 |
| Maxillary incisors | ⅔ to NA (%) | 22 | 25 | 30 | 30 |
| | ⅔ to NA (mm) | 4 | 5 | 7 | 7 |
| Mandibular incisors | ⅔ to NB (%) | 25 | 20 | 26 | 26 |
| | ⅔ to NB (mm) | 4 | 3 | 4 | 4 |
| | ⅔ to Md plane (%) | 92 | 95 | 105 | 105 |
| Interincisal angle (%) | 130 | 135 | 124 | 124 |
| Vertical | LFH/TFH (%) | 55 | 54 | 52 | 52 |
| Occlusal plane | SN-OP (%) | 14 | 14 | 12 | 12 |
| Soft tissue | Upper lip (mm) | -4 | -4 | -4 | -5 |
| | Lower lip (mm) | -2 | -4 | -3 | -3 |
| | Nasolabial angle (%) | 110 | 92 | 101 | 100 |
LONG-TERM FOLLOW-UP OF HEMIMANDIBULAR HYPERPLASIA

Figure 1a. Pretreatment photographs.

Figure 1b. Pretreatment study models.
results were negative for hyperplastic condylar growth. Therefore, the decision was made to proceed with comprehensive orthodontic treatment planning and care. The aetiology of HH is unknown; however, the literature currently supports a theory that describes an excessive response of the mandibular condylar cartilage to normal growth stimuli.²

**Treatment objectives**

The objectives of treatment were to achieve corrective goals facially, skeletally, and dentally in three planes of space: vertical, horizontal and transverse.⁵⁻⁷ Surgically levelling the projection of the maxilla and mandible would address the vertical facial asymmetry, vertical skeletal asymmetry, occlusal cant, and asymmetric dentogingival aesthetics. A clockwise rotation of the maxillomandibular complex during surgery would improve the hypodivergent skeletal pattern whilst enabling correction of the retrognathic maxilla, a reduction in overbite, and correction of the upper and lower incisal inclinations. A clockwise rotation of the occlusal plane during orthognathic surgery would also feminise the brachyfacial appearance. A combined surgical-orthodontic plan would align the dental arches and correct the dental midline discrepancy with differential dental and skeletal movements.
The restoration of the peg-shaped right lateral incisor to ideal proportions would correct the Bolton discrepancy, coupled with the combined assistance of interproximal reduction of the lower incisors. The interproximal reduction of the lower anterior teeth was also planned for the additional benefit of preventing orthodontic relapse.8,9

**Treatment alternatives**

Only a surgical-orthodontic treatment plan would achieve facial, skeletal and dental goals in the three planes of space. The surgical plan of bimaxillary surgery with a clockwise rotation of the occlusal plane was to be achieved by a Le Fort I osteotomy with differential superior repositioning of the maxilla on the right and left sides to correct the vertical maxillary asymmetry and frontal cant to the occlusal plane, and with advancement of the upper central incisor to rotate the occlusal plane clockwise from the lateral view. Secondly, a mandibular bi-sagittal split osteotomy was planned to correct the transverse mandibular asymmetry and dental midline discrepancy, match the lower occlusal plane to the upper, and decrease the overbite. Finally, a right mandibular inferior body osteotomy was considered to fully correct the vertical mandibular asymmetry and vertical facial asymmetry.

Orthodontic treatment without orthognathic surgery, including the use of skeletal anchorage devices such as mini-implants or bone-plates, may have corrected the occlusion initially; however, the facial and skeletal goals would not have been achieved. The patient’s face would have remained asymmetric and, therefore, compromised dentofacially from an aesthetic and a functional perspective. The use of skeletal anchorage to intrude the upper right posterior segment may have enabled an improvement in gingival display and correction of the occlusal cant. However, the long-term stability of intrusive dentoalveolar changes produced during orthodontic treatment with skeletal anchorage remains in question.10 Conversely, the excellent long-term stability of superior repositioning of the maxilla and bimaxillary surgery with a clockwise rotation of the maxillomandibular complex as a result of surgery is well documented.11-15

**Treatment progress**

Following a presentation of the orthognathic plan and obtaining informed consent, treatment was conducted in four stages: 1) pre-surgical orthodontic; 2) surgical; 3) post-surgical orthodontic; 4) post-treatment orthodontic retention.

Following the extraction of the upper right deciduous canine and restoration of the peg-shaped upper right lateral incisor to ideal proportions, the pre-surgical orthodontic treatment commenced with full-fixed appliance therapy, including placement of Burstone hinge-caps and the use of a 0.032” TMA transpalatal arch to control molar rotation and assist with dental alignment and levelling. Archwire selection included 0.020” × 0.020” nickel titanium to achieve vertical dental decompensation. Inter-arch correction was achieved in preparation for orthognathic surgery by the coordination of the upper and lower archwires using 0.019” × 0.025” stainless-steel wires with the mandibular arch form as the template.

The surgical stage included making complete pre-surgical records with clinical photographs, articulated study casts (using a facebow transfer of the patient in Natural Head Position16), cephalometric and panoramic radiographs, and a facial soft-tissue analysis. An immediate pre-surgical work-up included plaster model-surgery for surgical splint fabrication and computer-aided fabrication of a three-dimensional replica of the patient’s mandible to assist with special surgical planning.5 Bimaxillary surgery was planned to achieve: 1) the correction of the vertical facial asymmetry, 2) levelling of the vertical projection of the maxilla and mandible, 3) a clockwise (upwards and forwards) rotation of the occlusal plane and a reduction of the overbite, and 4) positioning of upper and lower dental midlines to the facial midline. An excellent surgical outcome was achieved and confirmed by immediate post-surgical radiographs and computed tomography. The surgical splint was maintained for a period of four weeks after surgery and used in combination with light (2oz, 5/16”) box-configuration elastics.

Post-surgical orthodontic treatment was completed five months after surgery, for a total treatment time of 30 months. A complete set of post-treatment records was taken at the time of debanding, which consisted of clinical photographs (extra-oral and intra-oral), study models, and cephalometric and panoramic radiographs. Interproximal reduction was performed at the time of debanding as planned. A period of positioner use, in combination with removable clear retainers, was to be reviewed after two months.
Following the period of positioner use, the patient continued full-time wear of the clear retainers for a period of six months, followed by night-time wear indefinitely. At 12-months post-treatment, the patient reported only intermittent retainer wear. An upper Hawley retainer was made and the patient continued night-time wear of upper and lower removable retainers. A passive Hawley retainer was used to assist with maintenance of the overbite correction. Regular clinical and radiographic orthodontic follow-up examinations were coordinated with the patient over a seven-year review period. The patient has required further endodontic and restorative treatment during orthodontic retention, and has elected to discontinue wearing orthodontic retainers.

**Treatment results**

Combined surgical-orthodontic treatment addressed the facial, skeletal and dental problems for this patient. The vertical facial asymmetry and vertical skeletal asymmetry were corrected. The chin was repositioned to the facial midline and a 100% improvement in the PAR Index was achieved. Post-treatment records are presented in Figure 3 (a-d). A mild Class III skeletal tendency was produced during the surgical stage. Factors contributing to this include the abnormal condylar morphology with associated difficulty of accurately positioning the condyle within the glenoid fossa during the orthognathic surgery, and the increased thickness of the surgical splint used. The Class III skeletal tendency may have contributed to the difficulty in closing the space distal to the upper left canine. During retention, orthodontic relapse occurred with rotation of the upper left first bicuspid and space opening between the upper canine and first bicuspid on the left side, which could have been prevented by the use of fixed retention and/or gingival fiberotomy of the bicuspid tooth.9,17 Despite these criticisms, the patient remains very satisfied with the facial, skeletal, and dental outcomes achieved during treatment.

**Figure 3a. Post-treatment photographs.**
Annual orthodontic follow-up visits were scheduled during the retention stage, with progress records taken at each visit, culminating with seven-year full orthodontic records obtained in 2015 (Figure 4a-d). Cephalometric superimpositions illustrate the dental, skeletal and soft tissue profile effects of the surgical-orthodontic treatment, and the excellent long-term stability of the results (Figure 5a-b). Minor postsurgical hard and soft tissue changes were evident during the post-treatment period, and are shown in Figure 5b. The long-term soft-tissue changes are likely due to maturational growth developments and postsurgical neuromuscular adaptation.18

Discussion

Hemimandibular hyperplasia (HH), also known as hemimandibular hypertrophy, is a rare condition characterised by excessive unilateral, three-dimensional growth of the mandible after birth, and, therefore, constitutes a true growth anomaly.1,2 The entire hemimandible (condyle, ramus, and body) is volumetrically enlarged to the mandibular midline symphysis, with the lower dental midline deviated towards the affected side. Patients affected by severe forms of HH have a frontal cant to the occlusal plane down towards the affected side and vertical facial asymmetry with concurrent compensatory vertical maxillary growth and excessive gingival display on the affected side.
Figure 4a. Seven-year post-treatment photographs.

Figure 4b. Seven-year post-treatment study models.
The severity of dentofacial deformity of HH necessitates combined surgical-orthodontic correction and care. The aetiology of HH remains largely unknown and an extensive discussion of the possible causes of HH is beyond the scope of this report; however, the current literature supports the theory of an excessive response of the mandibular condylar cartilage to normal growth stimuli. Hyperactive condylar growth translating to condylar hyperplasia is often a characteristic of HH, and condylar hypersensitivity to adolescent growth factors is a possible mechanism for initiating the excessive condylar response. However, the excessive growth at the condylar head does not explain the excessive three-dimensional growth of the entire hemimandible. Therefore, further aetiological mechanisms, as yet undiscovered, must be considered, which possibly include hypervascularisation, inflammation, and genetic factors. In addition, given the unknown aetiology of HH, the terms hemimandibular hypertrophy and hemimandibular hyperplasia are used interchangeably to describe the same clinical entity within the scientific literature. The condyle of HH may be characterised histologically by the appearance of mixed cartilaginous-bony trabeculae lacking...
in clear orientation, and remnants of cartilage matrix at abnormal distances from the erosion front of endochondral ossification. By comparison, the histological condition of the condyle affected by the hemimandibular elongation type of condylar hyperactivity anomaly may be characterised by well-oriented bony trabeculae, cartilage of normal structure and thickness, and subchondral cartilage rests at normal distances from the zone of erosion. The histological differences between the condyles of HH and the hemimandibular elongation type of condylar hyperactivity anomaly are suggestive of distinct pathogenetic processes; however, further comprehensive studies are needed to confirm these findings. Further research may achieve greater success in improving current knowledge of the aetiology of each type of condylar hyperactivity anomaly, and, therefore, identify potential improvements for treatment and management of their respective dentofacial deformities.

The excessive mandibular growth of HH may continue into the third and fourth decades of life. Clinicians must carefully and fully assess the patient before commencing comprehensive orthodontic treatment to ensure facial growth has ceased. Continuing assessment during orthodontic retention is imperative due to the potential for continued mandibular growth as a possible source of treatment relapse. Patients seeking surgical-orthodontic care to correct a dentofacial deformity caused by HH must be advised of the possibility of excessive mandibular growth into the retention phase of treatment, which should be monitored clinically and radiographically at annual intervals. Clinical examination, accurate study models, photographic records (extra-oral and intra-oral), and radiographic analysis are each equally important for the diagnosis of asymmetry, including HH, and for the assessment of orthodontic treatment relapse. Radiographic examination includes lateral and posteroanterior cephalograms, and three-dimensional imaging where possible and beneficial for diagnostic purposes. Serial lateral cephalometric superimpositions and radioisotopic (99mTc) bone imaging may confirm that craniofacial skeletal growth has ceased, to allow surgical-orthodontic treatment to proceed. Provided that the appropriate timing for surgical intervention has been determined through the use of serial lateral cephalograms and (99mTc) bone scanning modalities to confirm that facial growth has stopped.
ceased, patients and clinicians may be confident of the excellent long-term stability of a surgical-orthodontic plan to correct a HH. However, the possibility of continued mandibular growth is a source of future change and each case should be evaluated on an individual basis.

Historically, mandibular condylar hyperplasia was first reported by Adams in 1873, and Rushton further documented the unilateral predisposition of this condition, which included HH. However, the term condylar hyperactivity has since been defined by Obwegeser to explain mandibular asymmetries that are due to a surplus of growth activity affecting the entire hemimandible. These anomalies include HH and hemimandibular elongation (HE), and a rarer hybrid form of HH and HE, which are generally described in the literature under the term condylar hyperplasia. However, condylar hyperplasia is currently considered a generic term to describe excessive growth and/or enlargement of one or both of the mandibular condyles (not the entire hemimandible). Aetiological possibilities for condylar hyperplasia include neoplasia, trauma, infection, aberrant growth factors and idiopathic conditions. Varying degrees of condylar hyperplasia should be expected for condylar hyperactivity anomalies, and should be assessed independently using radioisotopic bone scanning before commencing treatment to ensure a thorough evaluation of each case. The results of bone scanning are highly sensitive but non-specific and are unable to determine the cause of the condylar hyperplasia or condylar hyperactivity. Patients experiencing condylar hyperactivity may benefit from a high condylectomy procedure to limit the effects of condylar overgrowth. However, given the potential for long-term three-dimensional growth beyond the somatic growth curve in cases of HH, further orthognathic surgery is likely to be needed once growth has ceased. Conversely, in the case of HE, excessive mandibular growth occurs along normal growth axes and is more closely timed with the somatic growth curve, therefore simultaneous condylar and orthognathic procedures are more likely to be successful. For the presented case of HH, cessation of hyperplastic condylar growth was confirmed by serial lateral cephalometric superimpositions and 99mTc bone scanning prior to commencing comprehensive surgical-orthodontic care, and there was no indication that supported a condylar surgical procedure during orthognathic surgery.

Clinical differentiation between the types of condylar hyperactivity is necessary for accurate diagnosis and comprehensive orthodontic care. Cases of HE are distinguishable from HH by their disproportionately excessive horizontal mandibular growth, deviation of the chin and dental midline to the unaffected side, a posterior crossbite on the unaffected side, a lack of frontal cant to the occlusal plane and a lack of vertical facial asymmetry. A diagnosis of the rarer hybrid form of HH and HE is more challenging, however, and is based on the facial appearance as a simultaneous combination of HH and HE with vertical facial asymmetry and deviation of the chin to the unaffected side. The pattern and severity of malocclusion of the hybrid variant type of condylar hyperactivity anomaly depends on the rate, timing, duration, and directions of excessive mandibular growth for each particular case. In severe cases, the lower dental arch may be rotated significantly out of occlusion.

Summary and conclusions
A severe case of hemimandibular hyperplasia is presented. Successful treatment was delivered by a combined surgical-orthodontic approach to correct a rare form of dentofacial deformity characterised by vertical facial asymmetry, vertical skeletal asymmetry with frontal canting to the occlusal plane, and a lateral open bite Class I-type malocclusion. A precisely considered and well-prepared surgical-orthodontic treatment plan will achieve correction of the dentofacial deformity caused by HH, and orthodontists, surgeons and patients may expect long-term stability of the results.

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