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Shin Pey Peng
Chang Gung Memorial Hospital, Taipei, Taiwan.

Wen-Ching Ellen Ko
Chang Gung Memorial Hospital, Taipei, Taiwan.

Cheng-Hui Lin
Chang Gung Memorial Hospital, Taipei, Taiwan.

Chung-Yi Ho
Chang Gung Memorial Hospital, Taipei, Taiwan.

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Abstract

Hemimandibular hyperplasia is a developmental asymmetry characterized by 3-dimensional enlargement in one half of the mandible. The malformation results in facial asymmetry, associated with tilted occlusal plane and a deviated chin to the contralateral side. These cause esthetic and progressive functional problems and need to be corrected without recurrence.

This report describes a 27-year-old woman presented with skeletal Class III malocclusion, occlusal plane cant and facial asymmetry. Radiographic films showed hyperplasia of half mandible and condyle on the left side. The condylar hyperactivity over left mandibular condyle was also confirmed by 99m-Tc bone scan. In order to correct her skeletal deformity, LeFort I osteotomy was performed to correct the secondary occlusal plane canting; extraoral vertical ramus osteotomy and condylectomy were performed in the left mandible, and sagittal split osteotomy was performed in the right mandible. Sliding genioplasty was also done to shift the chin in line with the facial midline. Good esthetic outcome and functional occlusion were achieved after the 22 months of post-surgical orthodontic treatment.

Keywords

Hemimandibular hyperplasia; Class III malocclusion; Sagittal split osteotomy; Vertical ramus osteotomy; Condylectomy

CASE REPORT

Hemimandibular Hyperplasia Treated by Orthognathic Surgery and Condylectomy in a Young Woman

Shin-Pey Peng [a,b], Chung-Yi Ho [a,c], Cheng-Hui Lin [d,e], Ellen Wen-Ching Ko [b,c,d,*]

[a] Department of Craniofacial Orthodontics, Chang Gung Memorial Hospital, Taoyuan, Taiwan
[b] Graduate Institute of Dental and Craniofacial Science, Chang Gung University, Taoyuan, Taiwan
[c] Department of Craniofacial Orthodontics, Chang Gung Memorial Hospital, Taipei, Taiwan
[d] Department of Plastic and Reconstructive Surgery, Chang Gung Memorial Hospital, Taoyuan, Taiwan
[e] Craniofacial Research Center, Chang Gung Memorial Hospital, Linkou, Taiwan

ABSTRACT

Hemimandibular hyperplasia is a developmental asymmetry characterized by 3-dimensional enlargement in one half of the mandible. The malformation results in facial asymmetry, associated with tilted occlusal plane and a deviated chin to the contralateral side. These cause esthetic and progressive functional problems and need to be corrected without recurrence.

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Keywords: Hemimandibular hyperplasia; Class III malocclusion; Sagittal split osteotomy; Vertical ramus osteotomy; Condylectomy

INTRODUCTION

Hemimandibular hyperplasia (HH) is a developmental asymmetry characterized by 3-dimensional enlargement in one half of the mandible. The malformation results in facial asymmetry, associated with tilted occlusal plane and a deviated chin to the contralateral side. These cause esthetic and progressive functional problems and need to be corrected without recurrence.

In HH, the hyperplastic side usually involves the condyle, condylar neck, ramus and body, with the anomaly terminating at the symphysis. The malformation results in the clinical presentation of ipsilateral enlargement of the mandible and tilted occlusal plane, associated with a deviated chin to the contralateral side.[1−3] The etiology of HH was originated from the factors associated with genetics, trauma, hormone, functional disturbance or tumor, but was still not clarified and well concluded. A previous study presented that it is related to osteochondroma.[4] It had greater prevalence in women.[5] It may also cause temporomandibular joint (TMJ) dysfunction such as clicking sound and pain due to the condylar head hyperplasia and malocclusion. HH can be diagnosed through clinical

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* Corresponding author at: Department of Craniofacial Orthodontics, Chang Gung Memorial Hospital, 6F, 199, Tung Hwa North Road, Taipei 105, Taiwan. Fax: +886 2 25148246.
E-mail address: ellenko.wc@msa.hinet.net (E.W.-C. Ko).

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manifestation, radiographic examination and bone scanning. Early diagnosis is really important because the treatment outcome depends on the affected structures and patient's age, severity of the asymmetry and the progressive status of the pathology. The diagnosis HH usually found in patients during young adult or adolescence.[6] Orthognathic surgery (OGS) combined with simultaneous condylectomy can produce functional and esthetic results.[7] We presented a case with HH with skeletal Class III malocclusion and elaborate the diagnosis and progress of the surgical-orthodontic treatment.

CASE REPORT

This 27-year-old woman came to our department with a chief complaint of facial asymmetry and crooked teeth. She denied any major systemic disease and history of TMJ disorders.

Clinically, right side up occlusal plane cant, chin deviated 5.5 mm to her right side and larger left face were noted (Figure 1). Intraorally, 0.5 mm overbite and overjet, Angle Class III malocclusion on both sides with posterior lingual crossbite on the right side were presented (Figure 2). Teeth 17, 18, 28 were extracted (missing) due to unrestorable tooth structure. Teeth 21−22 were splinted protheses. The 25−27 was a bridge with 26 missing. Teeth 14, 41 were crowns with fair condition. The vertical asymmetry of the maxilla and mandible was also confirmed in the PA cephalogram and two lines projection of the mandibular lower border in the lateral cephalogram (Figure 3). The enlarged left mandibular half in condyle as well as mandibular ramus and body was presented in the panoramic film. The inferior alveolar nerve was lower positioning in the left side. The cephalometric analysis revealed that the underlying skeletal pattern was hyperdivergent with mild skeletal Class III malocclusion characterized by an orthognathic maxilla and a mild prognathic mandible (Table 1). The condylar hyperactivity was confirmed by the count ratio of 1.5 for radioactivity over the left TMJ to the sphenoid sella in 99m-Tc bone scan (Figure 4).

Diagnosis

1. Hemimandibular hyperplasia in left side.
2. Skeletal Class III jaw relationship with mandibular prognathism, facial asymmetry with chin to her right side by 5.5 mm.
3. Angle Class III malocclusion with 12−16 lingual crossbite.

Treatment objectives

The aims of the treatment for this patient were established as follows:

(1) Correct facial asymmetry and occlusal plane cant.
(2) Correct skeletal Class III jaw relationship.
(3) Correct 12−16 lingual crossbite and achieve Class I canine and molar relationships.
(3) Remove the hopeless teeth and maintain 26 space for prothesis fabrication.

After discussion with the patient, the plan of 2-jaw surgical-orthodontics with surgery-first approach was set up as follows:

1. General dental care: including full mouth scaling, removal of 25−27 fixed prosthesis and replaced by 25, 27 individual provisional prothesis, 13 endodontic treatment with provisional prothesis, 18, 17, 28, 38, 48 extraction.
2. Full mouth bonded with edgewise orthodontic appliance.
3. Two-jaw orthognathic: LeFort I osteotomy in the maxilla; extraoral vertical ramus osteotomy (EVRO) accompany with condylectomy on the left mandible, sagittal split osteotomy (SSO) on the right mandible.
4. Post-surgical orthodontic treatment: including upper and lower arch coordination, dental and facial midline coordination, finishing and detailing of the occlusion and dental alignment.

Figure 1. Pretreatment facial photographs.

Figure 2. Pretreatment intraoral photographs.

5. Retention: wraparound retainer on the upper arch and Hawley retainer on the lower arch.

Treatment progress

Full mouth was bonded with edgewise brackets in one month before the surgery. Presurgical records included clinical examination, dental models with centric occlusion registration and computed cone-beam tomography (CBCT). The STL file of dental models were obtained by 3D scan to replace the denture parts in the CBCT, which commonly presented image scattering in 3D reconstruction. The surgical occlusion setup and simulated osteotomies were performed in 3D surgical simulation. By maxilla LeFort I osteotomy, the cant of the occlusal plane was corrected with roll rotation. To setback the mandible, SSO on the right side and EVRO on the left side were performed. The left condylectomy allow the lower part of left osteotomized ramus moving upward to replace and mimic the condylar head by contouring the upper bone edge. The left mandibular disc was preserved intact and kept in original position. Genioplasty was also done to shift the chin in line with the facial midline. Left mandibular inferior border contouring was performed to reduce the vertical overgrowth of the mandible (Figures 5 and 6). Preservation of the left inferior alveolar nerve was carefully monitored during surgery. Rigid internal fixation by screws and miniplates were placed both in the maxilla and mandible. Postsurgical orthodontic treatment was completed in 22 months after OGS. A complete set of posttreatment records was collected after fixed

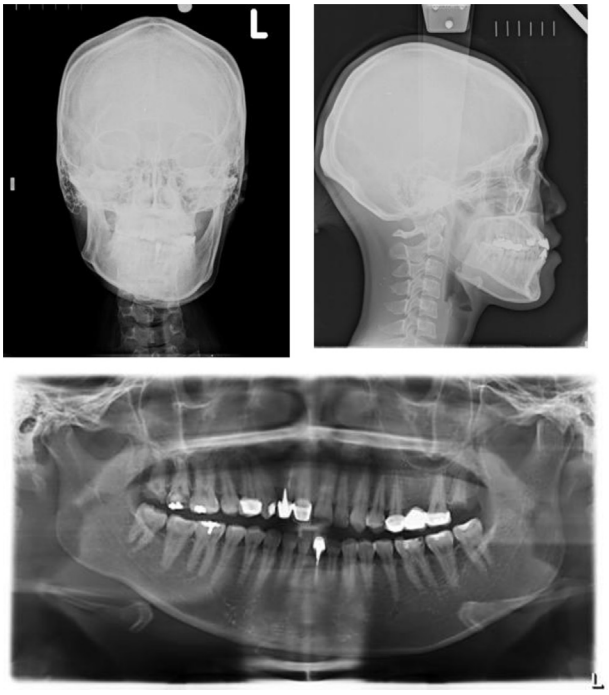

Figure 3. Pretreatment PA, lateral cephalometric and panoramic radiographs.

Table 1. Cephalomsetric analysis before surgery and after debond.

Cephalometric measurement	Initial	Finished	Norm
SNA (°)	76.5	77.0	79.4–82.5
SNB (°)	78.0	76.0	74.6–77.8
ANB (°)	−1.5	1.0	4.1–5.7
SN-MP (°)	45.0	42.0	34.2–38.6
Dental	Initial	Finished	Norm
U1-SN (°)	110.0	112.5	103.5–109.1
U1-NA (mm)	8.5	9.0	3.8–7.2
L1-MP (°)	84.0	92.0	93.4–99.2
L1-NB (mm)	5.0	6.5	5.4–10.2
Soft Tissue	Initial	Finished	Norm
U lip to E-line (mm)	0	0.5	0.8–3.2
L lip to E-line (mm)	2.0	0.5	1.2–4.4

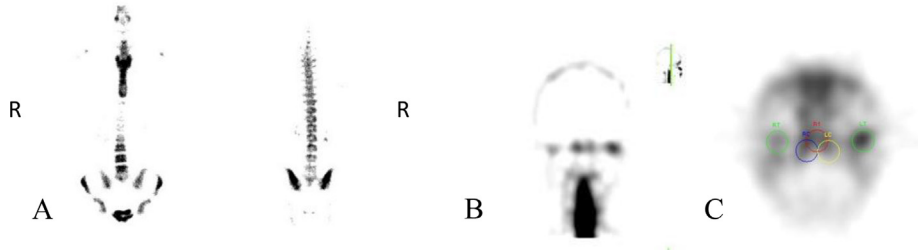

Figure 4. The 99m-Tc bone scan indicated condylar hyperactivity. A, Whole body bone scan; B, coronal view; C, axial view; the count ratio of radioactivity over the left TMJ to the sphenoid sella was 1.5.

appliance removal. The removable retainers were delivered to the patient for dental stability.

Treatment results

After a total of 23 months of surgical-orthodontic treatment, her facial asymmetry and occlusal plane cant were corrected (Figure 7). Bilateral mild Class II relationship and acceptable occlusal interdigitation was established; the transverse discrepancy and lingual crossbite was corrected by dental movement (Figures 8 and 9). The space of 26 was preserved for prosthesis. Resin teeth were placed on the upper removable retainer to keep the 17, 26 space for full-time wearing before definitive prosthesis. The new prosthesis would be replaced at least 3 months after debond. The constructed left mandibular condyle was remodeled to a normal joint contour and functioning well. The overall superimposition indicated upward movement in maxillary posterior occlusal plane and upward movement of the menton by 4.5 mm (Figure 10). There was a substantial vertical reduction and improvement in mandible border symmetry as well. For the soft tissue, her upper lip moved forward and labiomental fold was accentuated, resulting in a balanced lip posture. The cephalometric measurements presented that the SNB angle decreased by 2° and the SN-MP angle

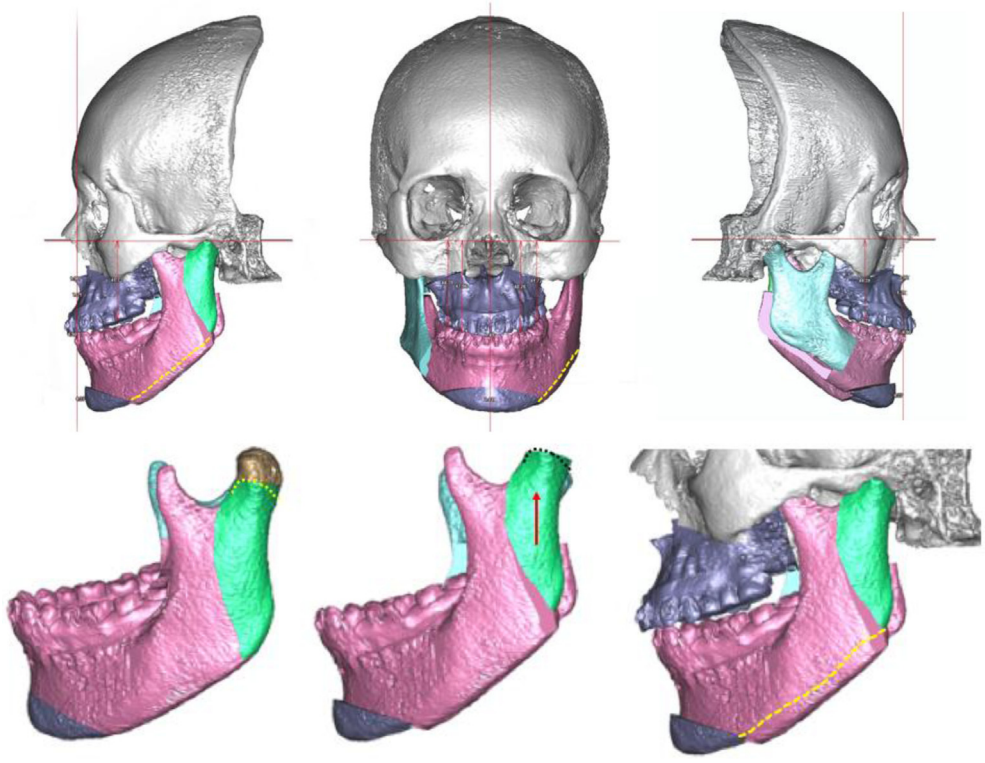

Figure 5. Surgical simulation. Included LeFort I can't correction, mandible setback 6 mm at the right side by SSO; condylectomy at the left mandibular condyle; VRO and upward movement to replace the condylar head; sliding genioplasty for better chin contour symmetry and projection; mandible border contouring was planned on the left side.

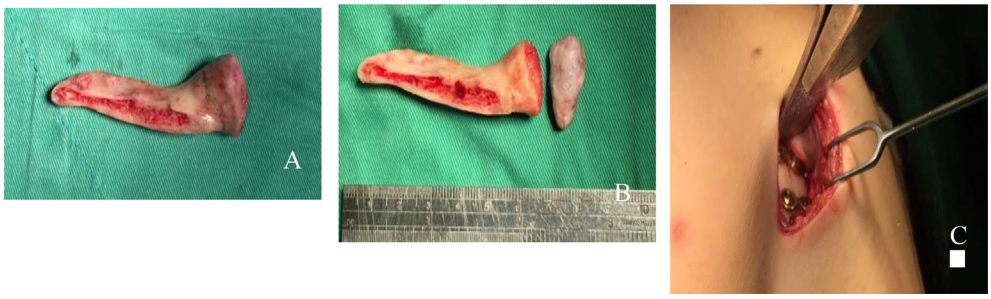

Figure 6. A, The excision of ramus/condyle unit was performed by VRO; B, the condylectomy was performed extraorally; C, the contoured ramus unit was placed back and secured with rigid fixation.

decreased by 3° (Table 1). The improvement of skeletal discrepancy of Class III relationship and anterior facial height was noted. The major changes of mandibular contour symmetry were demonstrated in 3D image superimposition, indicating a significant improvement of symmetry in transverse and vertical mandible borders (Figure 11). For mandible function, the impact on joint stability was limited. However, the mandibular range of motion was affected in lateral excusive movement toward her left side with normal maximum mouth opening at the time of debond.

DISCUSSION

For adult patients, correction of the asymmetries usually requires a series of complex surgical procedures. Therefore, diagnosis of the morphologic pattern of deficient or excessive growth is extremely important because it completely influences the orthodontic and surgical treatment outcome.

Typical HH is characterized by 3D enlargement in one side of the mandible, including enlargement of the condyle, condylar neck, ascending and horizontal ramus, as well as mandibular body.[1] Clinically, the increase in the vertical height of the middle and lower facial thirds on the affected side and downward-bowed mandibular lower border are important clinical traits. This should be distinguished from hemimandibular elongation (HE) and condylar hyperplasia (CH). Some controversies exist regarding the nomenclature of HH, HE and CH. However, according to study of Obwegeser, the main feature of HH is bone production in mass. As a result, the most striking features are deviation of the chin and the lower midline to the opposite side and a crossbite on the normal side.[3] We can also find double mandibular planes projection in the lateral cephalogram of patients with HH. There is substantial vertical height difference between the two sides of the face in HH, in contrast to HE. HE is mainly related with horizontal growth of mandible.[8] CH, which is so-called solitary condylar hyperplasia (SCH), is characterized by an enlargement of the condylar head or neck without the involvement of the ramus and the body. HH and CH are characterized with hyperplastic condyles with or without condylar deformation. Surgical excision could be considered for severely deformed condyles in HH or CH, whereas surgical excision is not necessary for HE.[9]

There are several methods that could complement the clinical and physical examinations, including radiographic analysis, extraoral photographs and photograph analysis for diagnosis of craniofacial asymmetries. Detail evaluation in frontal face in vertical and horizontal facial proportion is important.[8,10] Study models could also provide as a practical tool for the assessment of dental compensation, arch discrepancy and arch compatibility.[11] In addition to the conventional X-ray films, computed tomography now play an important role to evaluate the 3D structures in details. The simulation of 3D surgical changes enhances the surgical outcome and predictability.

As mentioned before, to deal with HH, evaluation of bone activity is important. The TC-99m scintigraphy should be assessed before the surgery to detect the mandibular bone and condyle activity.[12] It is a method which indicates the stability or activity of the hyperplastic condyle by comparison of cellular metabolic activity between normal and abnormal condyles.[13] The SPECT is a variation of scintigraphy and a method of functional imaging, which reflects the metabolic response of an organ.[14] The quantitative evaluation of radiopharmaceutical absorption at the condylar level in cases which are suspected as HH can be explored by comparatively detecting the percentage uptake between the two mandibular condyles, with reference to the activity in lumber spine or the clivus due to assumed symmetrical uptake.[15] As previous study investigated,

Figure 7. Posttreatment facial photographs.

Figure 8. Posttreatment intraoral photographs.

more than 10% difference in absorption between both condyles indicate one side condyle is more active and may lead to asymmetric growth.[8,14,16] In adult patients, if condylar hyperplasia is inactive, OGS alone is indicated for skeletal correction. If active condylar growth still presents, condylectomy is indicated together with standard OGS. The aim is to remove the joint cartilage that is responsible for active unilateral mandible growth.[17] A recent study found that after condylectomy, patients could still have good condylar function if they follow the postoperative physiotherapy plan.[18]

Surgical correction of HH can be conducted by different surgical techniques, which provide the solutions to address the patient's facial, skeletal, and dental problems. Both VRO and SSO techniques are well-known surgical methods to correct the mandible position.[2,3,10,19] Jensen et al. described a combination of a conventional SSO with a modification of this technique, which includes dissection of the neurovascular bundle from the mandible, horizontal intermediate bone reduction of the proximal fragment, and vertical reduction of the lower border of the distal fragment.[2] VRO is also indicated for hemifacial microsomia and correction of facial deformity which can be performed by either an EVRO or intraoral (IVRO) approach.[20]

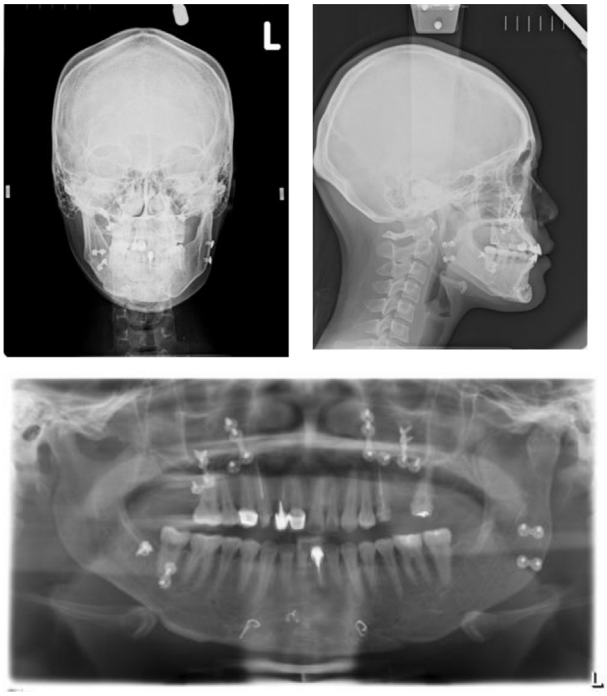

Figure 9. Posttreatment PA and lateral cephalometric and panoramic radiographs. The reconstructed left mandibular condyle was well remodeled.

Figure 10. Lateral cephalometric superimposition.

IVRO has the advantage of causing less frequent neurosensory disturbances without implicating scars.[21] However, it need 6 weeks of intermaxillary fixation because rigid internal fixation could not performed well in IVRO. In contrast, EVRO has advantages of direct access and good visibility as compared to IVRO. The technical difficulty of EVRO is preservation of inferior alveolar nerve and vascular bundle. Cautious pre-operative planning to confirm the location and orientation of nerve and vascular path is mendatary.[22] The anti-lingula is a common surgical landmark to guide the access of inferior alveolar nerve.[23] Surgical template or 3D printing guide is an effective method to increase surgical precision and safety in performing EVRO.[24]

In the present case, we used EVRO on the left side since we have to remove about 10 mm thick of condylar head.[25] The upper end of left osteotomized ramus was shaped to mimic the condylar head. The osteotomized ramus segment was moved upward to approach the condylar fossa and securely fixation with plate and screws to the mandibular body (distal segment). The mandible lower border contouring was performed after rigid fixation of EVRO. Attention should be paid to protect the inferior alveolar nerve during mandibular contouring. The line of mandibular border contouring should keep a distance below the inferior alveolar canal.[26] Thus, the amount of mandible border contouring has its limitation. With titanium miniplate fixation, it appeared to be minimal morbidity and few post-operative complications by combining EVRO and mandible border ostectomy. Rigid osteosynthesis with miniplates improve postoperative skeletal stability, avoid several weeks of intermaxillary fixation and early resumption of oral function for better convenience of the patients.[21] Functional rehabilitation of the jaw movement could be resumed earlier in one month after OGS.[27]

There was still some deficiency for orthodontic treatment outcome in this case. The occlusion was finished in mild Class II relation due to larger anterior Bolton ratio. The improvement should replace 11−12 splinted prothesis and increase the size of upper front tooth to compensate the discrepancy. In SFA, the postsurgical orthodontic treatment still spends 22 months. For this case, although we did SFA is for correction of mandible asymmetry and condyle pathology at earlier stage of treatment. And after the surgery, the orthodontic treatment was deferred for fully TMJ rehabilitation and stability. In addition, lingual crossbite and transverse discrepancy was corrected by dental movement after surgery instead of surgical expansion. The detailing still takes times to finish the final occlusion.

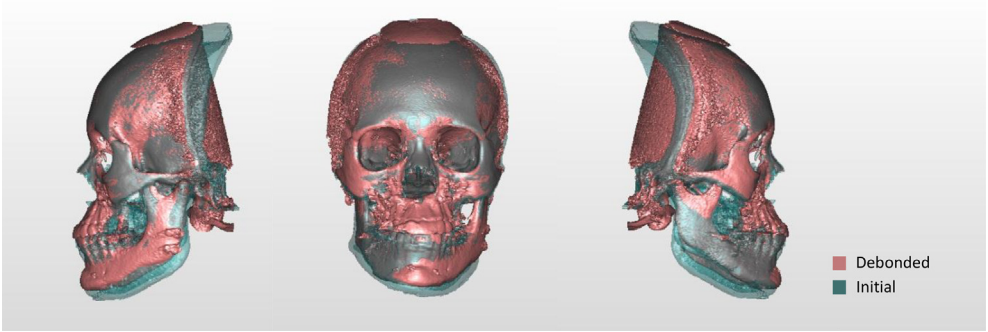

Figure 11. 3D images superimposition. Green color represents the image before treatment, red color represents the image at debond.

CONCLUSION

HH can be corrected by conservative to radical surgical procedure that base on the disease activity and severity of facial deformities. The extent of structural abnormality and the patient's expectations should be taken into consideration for choosing appropriate surgical techniques. In this case, with proper diagnosis and treatment plan, the abnormality and pathologic origin has been surgically corrected with fair aesthetic, functional results and minimal morbidity.

ETHICAL APPROVAL

This study was approved by the Institutional Review Board of Chang Gung Memorial Hospital. IRB: 202100408B0.

Conflict of Interest Statement

The authors declare no conflicts of interest.

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