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The accuracy of virtual-surgical-planning-assisted treatment of hemifacial microsomia in adult patients: distraction osteogenesis vs. orthognathic surgery

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Abstract. Hemifacial microsomia (HFM) is a common congenital craniofacial deformity with a high prevalence. Orthognathic surgery and distraction osteogenesis are two conventionally used treatments of HFM. The main objective of this retrospective study was to evaluate the accuracy of two treatments with the help of virtual surgical planning in adult HFM patients. Sixty-eight adult patients with unilateral HFM were enrolled in this study. Preoperative surgical planning and simulation were performed on three-dimensional computed tomography models. Orthognathic surgery or distraction osteogenesis was performed under the guidance of three-dimensional surgical templates. Postoperative evaluation of the intervention was performed by comparison of the affected ramus height, chin deviation and the occlusal cant in surgical planning and actual result. Outcome and feedback information (an average of 14 months) showed that virtual surgical planning was accurately transferred to actual surgery in both surgical approaches. There were no statistical differences between the accuracy of affected ramus height and the occlusal cant in two surgical approaches. The orthognathic group showed significantly higher accuracy in chin deviation. In conclusion, virtual surgical planning and three-dimensional surgical templates were proved to facilitate treatment planning and offer an accurate surgical result in the treatment of adult HFM patients.

Key words: hemifacial microsomia; orthognathic surgery; distraction osteogenesis; virtual surgery planning; 3D printing.

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Hemifacial microsomia (HFM), also called the syndrome of the first and second branchial arch, is the second most common congenital craniofacial malformation. The main characteristics of HFM include mandibular hypoplasia, facial asymmetry, unilateral or bilateral microtia with or without involvement of other facial structures. Due to the variety and complexity nature of HFM, its treatment remains a challenge even for the most experienced surgeons. Different surgical approaches including orthognathic surgery (OS) with bone graft and distraction osteogenesis (DO) were reported to correct the deformities of hard tissues and facial contour defects[1–3].

In the past, the accuracy of surgical procedures depended mainly on conventional model surgery and surgeons' clinical experience instead of quantitative analysis. Surgical templates fabricated from the model surgery guided the maxillary position. However, surgical planning procedure based on traditional plaster cast and surgical templates not only resulted in a variety of inaccurate consequences but also required a number of complicated laboratory steps including face bow transfer, mounting of a dental cast on the articulators and model surgery procedures[4]. For these reasons, virtual surgical planning (VSP) and the three-dimensional (3D) printing technique were developed to provide a more proper and accurate treatment for complex HFM[5–7]. VSP enabled the surgeons to make an accurate diagnosis, guided accurate surgical execution, and facilitated the analysis of postoperative tissue changes. Multiple studies have analysed the accuracy of VSP by comparing the preoperative surgical design and postoperative surgical results[8–10]. However, no study compared OS and DO in HFM patients with the aim of analysing the accuracy. Therefore, the aim of this study was to evaluate the surgical accuracy of VSP-assisted OS and DO in cases of adult HFM. Surgical splints were fabricated by rapid prototyping technology. The actual results of the surgeries were compared with the preoperational surgical plan by a 3D analysing method.

Patients and methods

From January 2007 to January 2016, 68 HFM patients (36 women and 32 men) were referred to our department (Center of Orthognathic and TMJ Surgery, West China College of Stomatology, Sichuan University, Chengdu, China). Ages ranged from 18 to 34 years and the follow-up was 12–24 months. All of them were categorized as Pruzansky–Kaban type I (20 patients) or type II (26 patients IIa, 22 patients IIb). Nearly all patients presented with facial deformities including micrognathia, occlusal cant, chin deviation, condylar dysplasia, shortened mandibular body on the affected side and facial asymmetry. Patients recorded with systemic diseases other than auditory or ocular system abnormalities caused by HFM or psychological disorders were excluded from this study. Patients whose facial deformities were due to acquired factors were also excluded. These patients were then separated into two groups according to the treatment procedures. Group I included patients who accepted OS, such as

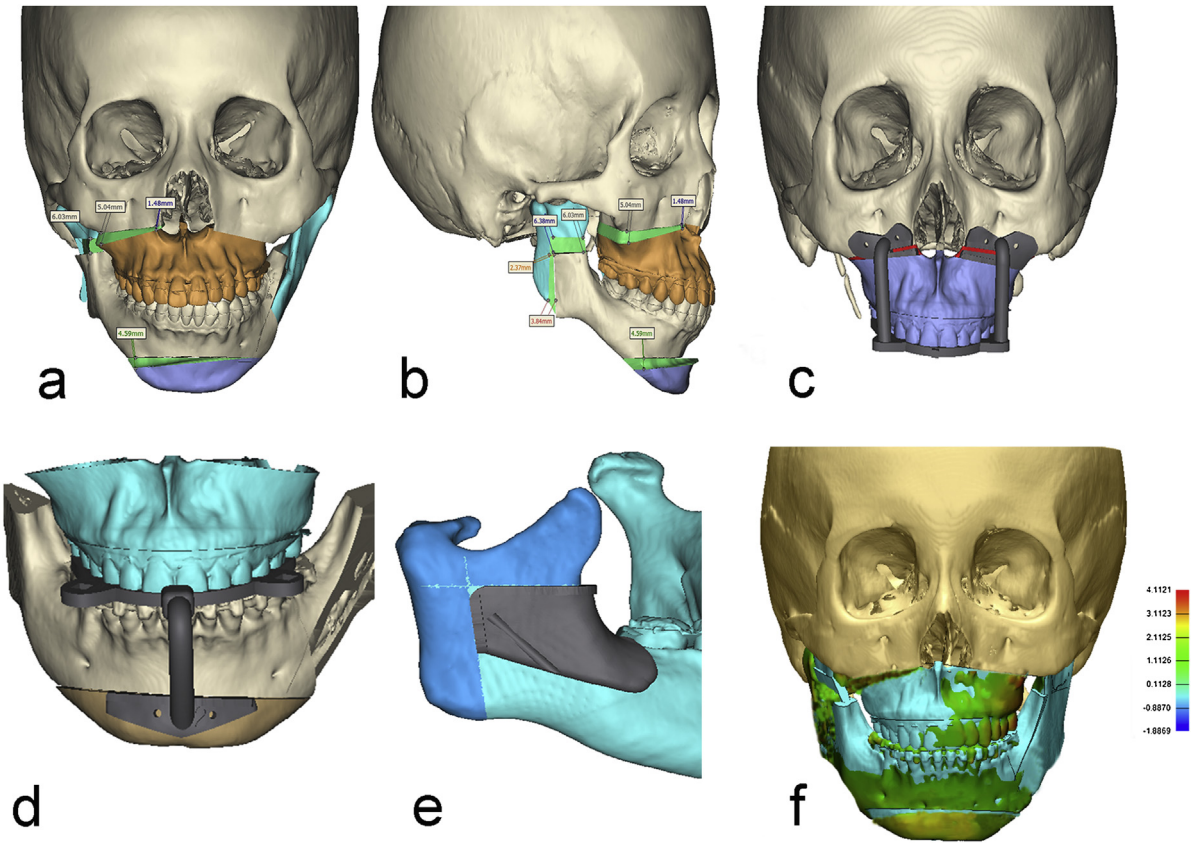

Fig. 1. (A, B) Illustration of design lines and procedures of maxillary Le Fort I osteotomy, inverted L osteotomy and genioplasty, sagittal split ramus osteotomy (SSRO) as well as iliac bone graft (A: frontal view; B: lateral view). (C–E) Illustration of simulated surgery and the design of the surgical templates. (F) Superimposed computed tomography images of virtually simulated and postoperative images. Metric differences of the surface for the bone tissue were displayed in the form of a colour scale.

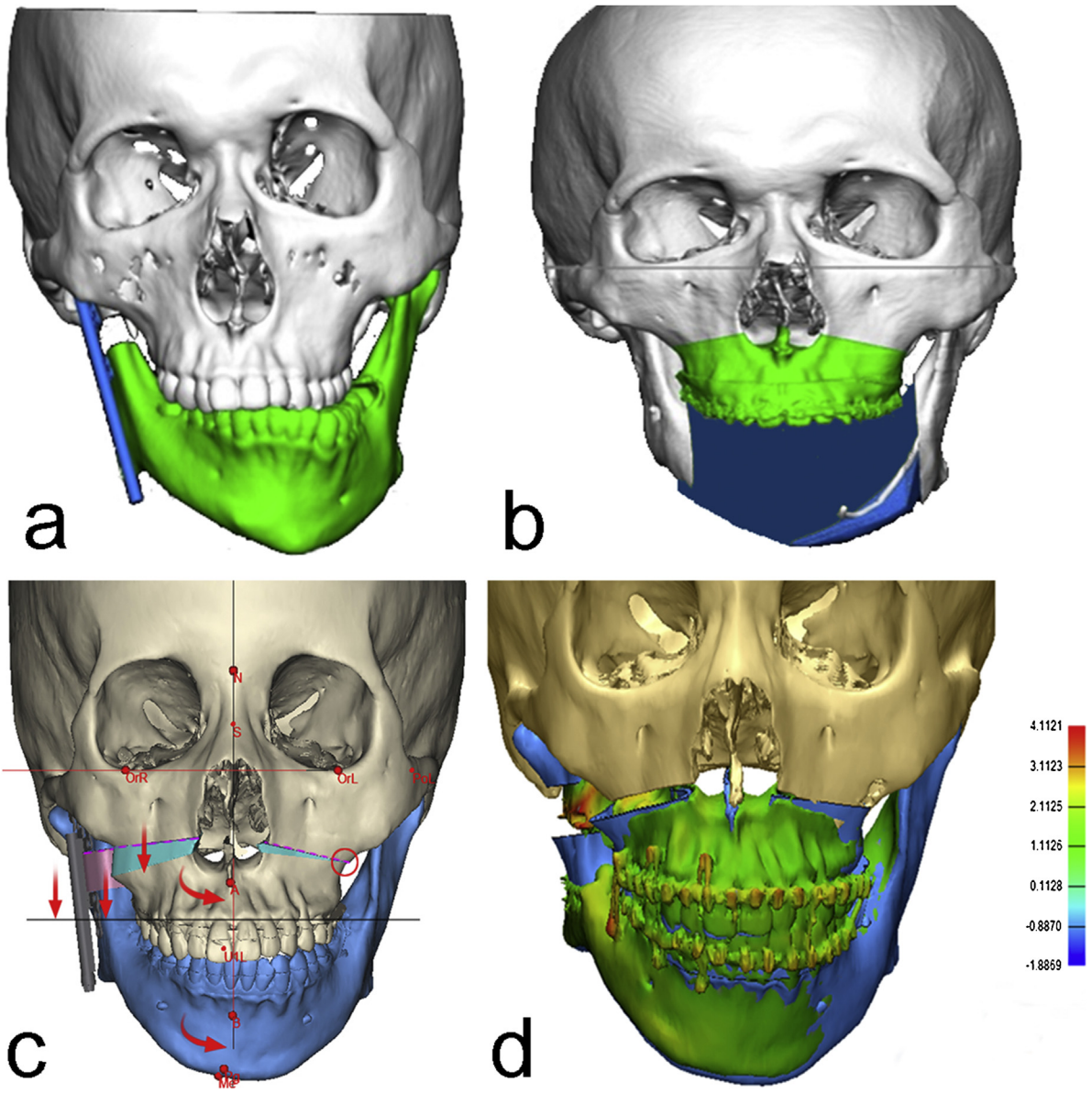

Fig. 2. (A, B) Illustration of designed lines of single mandibular distraction osteogenesis. (A) Position of the mandibular distractor. (B) Position of maxillary osteotomy line. (C) Illustration of design lines and procedures of bimaxillary distraction osteogenesis. (D) Superimposed computed tomography images of virtually simulated and postoperative images. Metric differences of the surface for the bone tissue were displayed in the form of a colour scale.

maxillary Le Fort I osteotomy, sagittal split ramus osteotomy (SSRO), genioplasty and invert L osteotomy on the affected side with bone graft. Group II included those who underwent DOboth single mandibular DO and bi-maxillary DO were included. There were five patients in the OS group with poor mandibular inferior border contour. High-density porous polyethylene (Medpor®, Porex Surgical Inc., Newman, GA, USA) was used at the second stage after postoperative measurement to repair mandibular contour deficiency. And thus Medpor® did not affect the result of the postoperative measurement. Orthodontic treatment pre- and postoperatively was commonly used in patients with severe occlusal discrepancy. Soft tissue correction such as vascularized flap implantation, free fat filling or prosthetic chin augmentation was also recommended for patients after skeletal symmetry was achieved.

Spiral computed tomography (CT) was conducted pre- and postoperatively. The CT images were obtained in Digital Imaging and Communications in Medicine (DICOM) format. Surface scanning of the dental arch was obtained in step ladder instruction (STL) format 1 month before the surgery. All the data were processed with Mimics software version 12.0 to construct a composite skull model with accurate dentition. Virtual surgery simulation was performed by Shenzhen PTY Medtech Co., Ltd. (Figs 1 A, B, 2 A–C). To provide an accurate presurgical evaluation and treatment plan for surgeons, 3D-printed surgical templates were fabricated using data from VSP by rapid prototyping technology (Fig. 1C–E). Besides, quantitative analysis of the movement of bone segments and the shape of transplanted bone provided a definitive guidance for the surgery. All surgeries were performed intraorally or extraorally under general anaesthesia through nasotracheal intuba-

Table 1. The difference between virtual surgery planning and postoperative results.

Surgical treatment	Type	Patients (*n*)	Affected-side ramus height (mm)				Chin deviation (mm)				Occlusal cant degrees (°)			
			T0 (mean ± SD)	T1 (mean ± SD)	|T0-T1| (mean)	*P*	T0 (mean ± SD)	T1 (mean ± SD)	|T0-T1| (mean)	*P*	T0 (mean ± SD)	T1 (mean ± SD)	|T0-T1| (mean)	*P*
OS	I	20	47.18 ± 4.52	48.45 ± 5.18	1.27	0.414	1.14 ± 0.28	1.02 ± 0.19	0.12	0.122	1.9 ± 0.3	1.8 ± 0.2	0.1	0.224
	IIa	16	46.12 ± 4.17	47.76 ± 4.55	1.64	0.296	1.31 ± 0.27	1.48 ± 0.36	0.17	0.141	1.1 ± 0.2	1.0 ± 0.1	0.1	0.087
	IIb	14	43.52 ± 4.28	45.18 ± 3.94	1.66	0.295	1.45 ± 0.26	1.32 ± 0.22	0.13	0.165	2.2 ± 0.4	2.0 ± 0.2	0.2	0.111
DO	IIa	10	46.08 ± 3.75	47.77 ± 4.88	1.69	0.397	2.05 ± 0.32	1.82 ± 0.27	0.23	0.099	2.6 ± 0.4	2.4 ± 0.3	0.2	0.222
	IIb	8	47.39 ± 5.12	46.26 ± 4.74	1.13	0.654	2.42 ± 0.63	2.10 ± 0.44	0.32	0.259	2.8 ± 0.5	2.9 ± 0.4	0.1	0.665

DO, distraction osteogenesis; OS, orthognathic surgery; SD, standard deviation; T0, VSP data 1 month before the surgery; T1, 1-year postoperative data; VSP, virtual surgery planning. Paired *t*-test was performed and no significant difference was found between VSP and actual surgery.

tion. Negative pressure drainage was used about 3 days postoperatively. Intermaxillary fixation (IMF) was performed in all patients using miniscrews or orthodontic appliances. Postoperative orthodontic treatment was followed after 3 weeks.

The difference between VSP 1 month before the surgery (T0) and 1−year postoperative results (T1) was measured in all the patients (Table 1). The differences between the OS and DO groups were also calculated (Table 2). The occlusal cant, ramus height and chin deviation were calculated, respectively, to evaluate the accuracy of VSP. Metric differences of the surface for the bone tissue were displayed in the form of a colour scale (Figs 1 F, 2 D). The affected-side ramus height was measured from the halfway point of the sigmoid notch to the inferior border of mandible through the lateral view of the CT image. The chin point deviation was measured using the distance between the chin point and the midline in CT frontal view. Furthermore, the postoperative analysis of occlusal cant was conducted by drawing a line between bilateral infraorbital margin and another line between the first molars of the upper jaw in the frontal view of the CT image. The angle between the two lines in the frontal view of the CT image represented the degree of occlusion cant (Fig. 3).

All medical practice followed the Declaration of Helsinki on medical protocol. This retrospective study was approved by the ethics committee of West China College of Stomatology and the institutional review board of Sichuan University. All participants signed an informed consent agreement.

Statistical Analysis

The methodical error of the cephalometric and facial measurements was assessed by Dahlberg's formula[11,12] (mean square error (standard error (SE)^2) = $d^2/2n$, where d is the difference between the first and the second measurements, and n is the number of double measurements). The VSP and postoperative data were analysed by paired (Table 1) and independent (Table 2) *t*-tests using the SPSS software (IBM SPSS Statistics for Windows, version 24.0 (IBM Corp, Armonk, NY, USA) to compare the differences. A *P*-value less than 0.05 was considered to be statistically significant.

Results

All patients tolerated surgery well and presented with satisfactory healing. The symptoms of facial asymmetry were also vastly improved. Table 1 presented the mean value and measurement differences between the VSP at 1 month before the surgery (T0) and actual 1-year postoperative results (T1) in both OS and DO groups. VSP was accurately transferred to the actual surgery based on the measurement data. The mean postoperative affected-side ramus height for OS and DO patients were 47.31mm and 47.10mm, respectively. The mean postoperative chin deviation for OS and DO patients were 1.25mm and 1.94mm, respectively. The mean postoperative occlusal cant for OS and DO patients were 1.6° and 2.6°, respectively. For the measurement difference, different Pruzansky–Kaban type was separately calculated. All the measured differences were within an acceptable range and there was no significant difference between VSP and actual surgery results in each Pruzansky–Kaban type ($P > 0.05$).VSP was accurately transferred to the actual surgery.

Table 2 presents the differences between the measurement data of the OS and DO groups. The mean measurement difference of affected-side ramus height in OS and DO groups was 1.50 mm and 1.44 mm, respectively. The mean measurement difference of chin deviation was 0.14 mm and 0.27 mm, respectively. And the mean measurement difference of occlusal cant degree was 0.13° and 0.15°, respectively. Although significant difference of the chin deviation was found between OS and DO groups ($P = 0.000$), no significant differences were seen in affected-side ramus height ($P = 0.363$) as well as occlusal cant degree ($P = 0.056$).

Table 2. Measurement differences analysis between two groups of virtual surgical planning and postoperative data results.

Measurement	Mean (OS)	Mean (DO)	SD (OS)	SD (DO)	*P*
Ramus height (%) (affected side)	1.50	1.44	0.32	0.20	0.363
Chin deviation (mm)	0.14	0.27	0.03	0.07	0.000
Occlusal cant degrees (°)	0.13	0.15	0.03	0.04	0.056

Sixty-eight patients were included. Independent *t*-tests were performed and significant differences were found in chin deviation between the OS and DO groups. DO, distraction osteogenesis; OS, orthognathic surgery; SD, standard deviation.

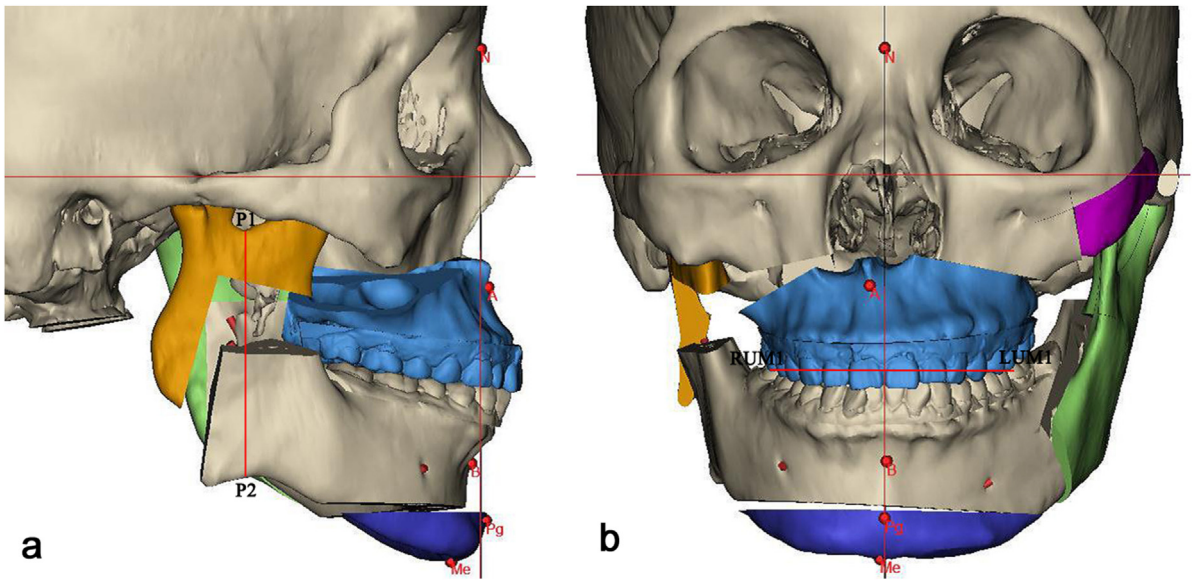

Fig. 3. (A) Measurement of affected-side ramus height in a lateral view of a computed tomography (CT) image. The distance between the halfway point of the sigmoid notch (P1) and inferior border of mandible (P2) represents the affected-side ramus height. (B) Measurement of chin deviation and occlusal cant in frontal view of CT image. The distance between the chin point (Me) and the midline represents the chin deviation. The angle formed by the line between bilateral infraorbital margin and another line between the first molars of the upper jaw (LUM1 and RUM1) represents the occlusal cant.

Discussion

The treatment for HFM relies on complex surgical planning with a thorough clinical examination and radiographic imaging. Conventional OS is used to solve a great variety of facial skeletal problems and restore the facial aesthetics and function in HFM patients[13]. Without bone graft and the second surgery area, DO is also used to solve bone volume deficiencies. It can lead to bone formation of high quality[14–17]. In addition, different kinds of soft tissue corrections should be considered only after all the required skeletal corrections have been completed[18–20]; this is especially critical for patients who undergo DO. The distraction alone facilitates mandibular vertical and anteroposterior lengthening but might not be able to address the 3D deformities sufficiently. The necessary soft tissue correction serves as the final surgical step in spite of some possible complications[21,22].

Most treatment outcomes of conventional different treatment procedures are still satisfactory, but due to the limitations of two-dimensional surgical planning methods for 3D deformities, less accuracy is shown in measurement, diagnosis and execution of various surgical procedures. In contrast, VSP based on 3D imaging yields more accurate anatomical information and enables the precise osteotomy and positioning of maxillary and mandibular segments[23]. VSP provides a cost-effective surgical planning method and reduces the operation time[24]. VSP also serves as an assistant to the whole treatment process and provides support from preoperative measurement and analysis to diagnosis and surgical design, intraoperative osteotomy, bone reposition, rotation and fixation. With regard to the accuracy, multiple studies analysed the accuracy of VSP by comparing the preoperative surgical design and postoperative surgical results in OS[8–10]. And VSP could be accurately transferred with the help of 3D-printed navigation templates in OS[25].

In this study, we focused on the comprehensive consideration and design with the help of VSP for treatment of HFM in the dentomaxillofacial deformities in adults. Our study provided a bridging methodology combining diagnosis and virtual planning to CAD/CAM and rapid prototyping technologies in treating HFM. We showed that this approach provided clinically acceptable precise transfer of preoperative VSP to the actual surgery for the affected-side mandibular ramus height (1.50 mm for OS and 1.44 mm for DO); chin deviation (0.14 mm for OS and 0.27 mm for DO) and occlusal cant degree (0.13° for OS and 0.15° for DO). As for the comparison of OS and DO, no significant difference was observed in affected-side ramus height and occlusal cant degree. Despite these results, we found that, statistically at least, chin deviation in the DO group was unable to be achieved with the same accuracy as in the OS group ($P = 0.000$). This difference is relative, however, and possibly of not of much clinical importance in actual deviations from planned to actual mandibular position. One possible source of error contributing to this relative imprecision might be the inaccurate actual distraction vector control. Determination of distraction vector remained a clinical challenge, especially in HFM patients whose distraction required not only lengthening, but also rotational movement of bones. Yu et al. reported the use of computer-assisted DO in the treatment of HFM and good matching with the preoperative vector design was achieved[26]. However, statistical error might occur in our own clinical experience.

Another shortcoming of this study was the lack of soft tissue simulation. VSP was unable to simulate soft tissue accurately and sometimes soft tissue remodelling disharmony could occur after hard tissue changes.

In conclusion, resulting from our retrospective study of 68 patients, we confirmed the clinical feasibility and accuracy of VSP-assisted treatment of HFM in adult patients using 3D-printed surgical templates. Further studies with larger sample sizes and soft tissue simulations are required to validate this technique.

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Competing interests

The authors have no conflicts of interest to declare.

Ethical approval

The authors read the Helsinki Declaration and followed the guidelines in this investigation. The research protocol was approved by the West China Hospital of Stomatology Institutional Review Board (WCSHIRB). The Judgement's reference number is WCHS-IRB-CT-2017-0136.

Patient consent

The patients involved in the article agreed to the use of photographs and patient consent forms were obtained.

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