Diagnostic Accuracy Study

Medicine®

OPEN

Three-dimensional printing of surgical guides for mandibular distraction osteogenesis in infancy

Zhe Mao, MD, Na Zhang, MD, Yingqiu Cui, MD*

Abstract

Pierre Robin sequence (PRS) is a congenital malformation characterized by micrognathia, glossocoma, and mechanical obstruction of the upper respiratory tract. These deformities impair respiration, sleep, feeding, and swallowing, and can lead to malnutrition, stunted development, and death. Bilateral mandibular distraction osteogenesis, whereby the mandible and tongue root are extended outward, is the standard treatment to relieve upper airway obstruction in severe PRS. Accurate placement of the distraction device is essential but challenging, especially in infants, and requires the pre-operative fabrication of surgical guides based on CT images. Three-dimensional (3D) printing allows for the accurate recreation of objects from digitized models. We compared surgical efficacy and safety of bilateral mandibular distraction osteogenesis using 3D printed or traditionally fabricated surgery guides for treatment of infants with severe PRS.

During the period from 2014 to 2016, 22 patients with severe PRS were treated using either traditional or 3D printed surgery guides. We compared outcome measures of operations, including intraoperative bleeding, operation time, and postoperative complications.

The 3D printed surgery guide group demonstrated significantly shorter operation time ($P<.05$) as well as moderately shorter hospital stay and artificial ventilation time (~1 day less). Furthermore, despite markedly younger average age of the 3D printed group (1.3 vs 3.5 months), there was no increase in postoperative complications using the 3D printed guides.

Three-dimensional printed surgery guides were used successfully for bilateral mandibular traction osteogenesis, and according to several outcome, parameters demonstrated superior efficacy and safety compared to traditional guides. Further research is warranted to extend the applications of 3D printed surgical guides for craniofacial surgery.

Abbreviations: 3D = three-dimensional, CT = computed tomography, PRS = Pierre Robin sequence.

Keywords: 3D print, bilateral mandibular traction osteogenesis, Pierre Robin sequence, surgery guide

1. Introduction

Pierre Robin sequence (PRS) is a congenital condition first described by the French physician Dr Pierre Robin in 1923.[1] PRS normally involves micrognathia, glossocoma, mechanical obstruction of the upper respiratory tract, and feeding difficulty due to these malformations. The estimated prevalence of PRS is 1/3000, and the associated mechanical obstruction of the upper respiratory tract and feeding difficulties can lead to secondary symptoms like respiratory and sleep disorder, malnutrition, slow to negative increase in body weight during development, and early death in severe cases.[2] For children with severe PRS, surgical treatment normally includes bilateral mandibular distraction osteogenesis in which the mandible is extended outward, thereby moving the tongue in the anterior and inferior directions and relieving the respiratory tract obstruction for improved respiratory function, sleep, and swallowing.[3] Infants suffering from PRS are usually small and poorly developed with severe malnutrition. Therefore, accuracy, reduced intraoperative bleeding, and short operation times are critical for outcome.[4] Thus, the pre-operational preparation of surgery guides is very important.[5] Most of the surgery guides used for mandibular distraction osteogenesis are hand-made, and so may carry disadvantages like poor dimensional accuracy, easy deformation, and long processing time, that do not meet the requirements of clinical use. Three-dimensional (3D) printing has several advantages for this purpose, including digitalization, flexibility, and customization during the fabrication process, all of which may improve dimensional accuracy and reduce complications. Indeed, 3D printed surgical guides are widely used in related fields, including fabrication of medical models for pre-surgical diagnosis and training,[6,7] customized implants, and medical devices,[8–10] and 3D printed cell/tissue models for drug testing.[11,12] Three-dimensional models customized to the diseased body parts of individual patients can provide surgeons with direct perception of the disease condition and facilitate the design of a better surgical plan. The application of the 3D printing technique has the potential to effectively reduce surgical risks and enhance accuracy and treatment efficacy.[13] In this study, the preparation of 3D printed surgery guides and use in

Editor: Yan Li.

The authors have no conflicts of interest to disclose.

Guangzhou Women and Children's Medical Center, Guangzhou Medical University, Guangzhou, Guangdong, China.

* Correspondence: Yingqiu Cui, Department of Oral and Maxillofacial Surgery, Guangzhou Women and Children's Medical Center, Guangzhou Medical University, Guangzhou, Guangdong 510120, P.R. China (e-mail: gzhtwang@163.com).

Medicine (2019) 98:10(e14754)

Received: 18 October 2018 / Received in final form: 1 February 2019 / Accepted: 9 February 2019

http://dx.doi.org/10.1097/MD.0000000000014754

Table 1

Preoperative patient characteristics.

Parameter	Control (n=12)	3D print (n=10)	*P* value
Birth weight, kg	2.9333±0.2987	2.8300±0.3093	.436, NS
Gestational age, wk	38.0833±1.5643	38.1000±1.5951	.981, NS
Age at operation, mo	3.5000±2.0671	1.2000±0.4216	.003, S
Males	9 (12)	5 (10)	.245, NS
Palatal cleft	10 (12)	10 (10)	.193, NS

NS=no significant difference ($P>.05$), S=significant difference ($P<.05$).

mandibular distraction osteogenesis are described and surgical outcomes compared to procedures using traditional manually fabricated surgery guides.

2. Patients and methods

2.1. Research patients

This non-randomized, retrospective single-center study included patients undergoing bilateral mandibular distraction osteogenesis for severe PRS disease at our hospital. Surgical options are mainly reserved for infants with severe airway obstruction who do not respond to non-invasive interventions aimed at mitigating the need for tracheostomy. The 22 patients were randomly divided into 2 groups, a 3D printed surgery guide group (3D print group) with 10 patients and a traditional surgery guide group (control group) with 12 patients. Before surgery, patient conditions were analyzed and compared (Table 1). This grouping method was approved by the ethics committee. The patient's consent was signed by parents or caregivers.

2.2. Preparation of surgery guides

All patients underwent computed tomography (CT) scanning before surgery. We transferred on mandibular structure to Mimics software for 3D modeling. For the control group, traditional surgery guides were prepared as follows. The 3D digital model of the mandible was transmitted to a 3D printer with STL data and printed. A wax model was handmade according to the printed model and the surgery guide was then made using bio-resin copying the wax model (Fig. 1). For the 3D print group, the 3D printed surgery guide was prepared as follows. A 3D model of the mandible was constructed by CT scanning data as described. Subsequently, the surgery guide model was designed by computer software and then fabricated of the same bio-resin using a 3D printer (Fig. 2).

2.3. Surgery procedures

All surgeries were performed by the same group of doctors following routine procedures; the only difference was the method of surgical guide preparation. The surgical guides were sterilized before surgical. The patient was placed horizontally in the supine position. Under general anesthesia, submandibular incisions were made to expose the bone surface, the surgery guide was placed on the bone surface and wrapped around the mandibular angle, and we performed osteotomy along the guide line. After osteotomy, a nonabsorbable device was implanted with a vector like this along the mid-point of the condyle to the chin point, parallel to the surface of the jaw. (Figs. 3 and 4)

2.4. Comparison of the surgery guide effect

Surgical outcomes and postoperative complications were compared between groups. Surgical outcome was assessed according to common indicators for PRS (Table 2) and common complications after bilateral mandibular traction osteogenesis (Table 3).

2.5. Statistical analysis

Statistical analyses were performed using Excel 14.3.9 and SPSS. Continuous variables are expressed as mean±standard error of the mean and categorical variables as proportions (%). Means of continuous variables were compared by the X test and proportions by the Y test. All tests were 2-tailed and a $P<.05$ was accepted as statistically significant.

3. Results

There were no significant differences in birth weight (kg), gestational age (week), ratio of males to females, and incidence of palatal cleft between the control group and the 3D print group (all $P>.05$) (Table 1). Of preoperative factors examined, only age at operation differed, with control patients significantly older ($P<.05$), indicating that average patient condition for surgery was actually superior in the control group.

During surgery, the traction process went well for both groups, the bone formed well at the surgical site, and no infections or other complications occurred in the perioperative period (6 months). Surgery efficacy and safety were rated according to 5 indices, surgery time (minutes), bleeding amount (mL), time of post-surgical separation of the artificial airway (days), time of hospitalization (days), and time of post-surgical oral feeding (days) (Table 2). Surgery time was significantly shorter for the 3D

Figure 1. Fabrication of a handmade surgical guide (control group). A) The mandible model is obtained. B) The handmade surgical guide is produced using self-condensing plastics. The osteotomy line and fixation screw positions are recorded. C) The final handmade surgical guide is obtained.

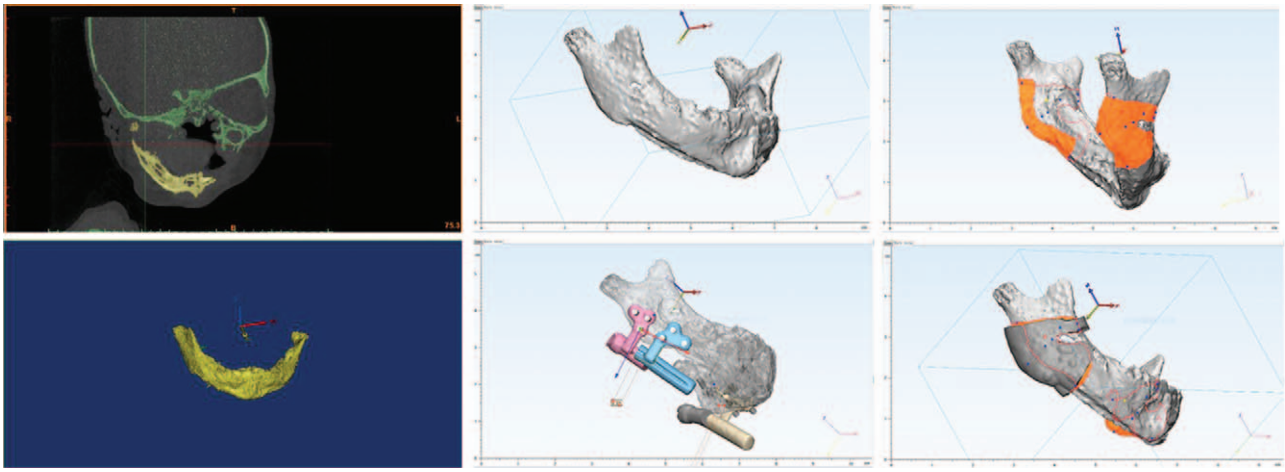

Figure 2. Fabrication of a 3D printed surgical guide. A) Data collection by CT. B) 3D modeling using Mimics software. C) Model import to 3magic software. D) The tractor position is simulated. E) The osteotomy line and fixation screw positions are determined. F) The surgical guide is designed and exported for 3D printing.

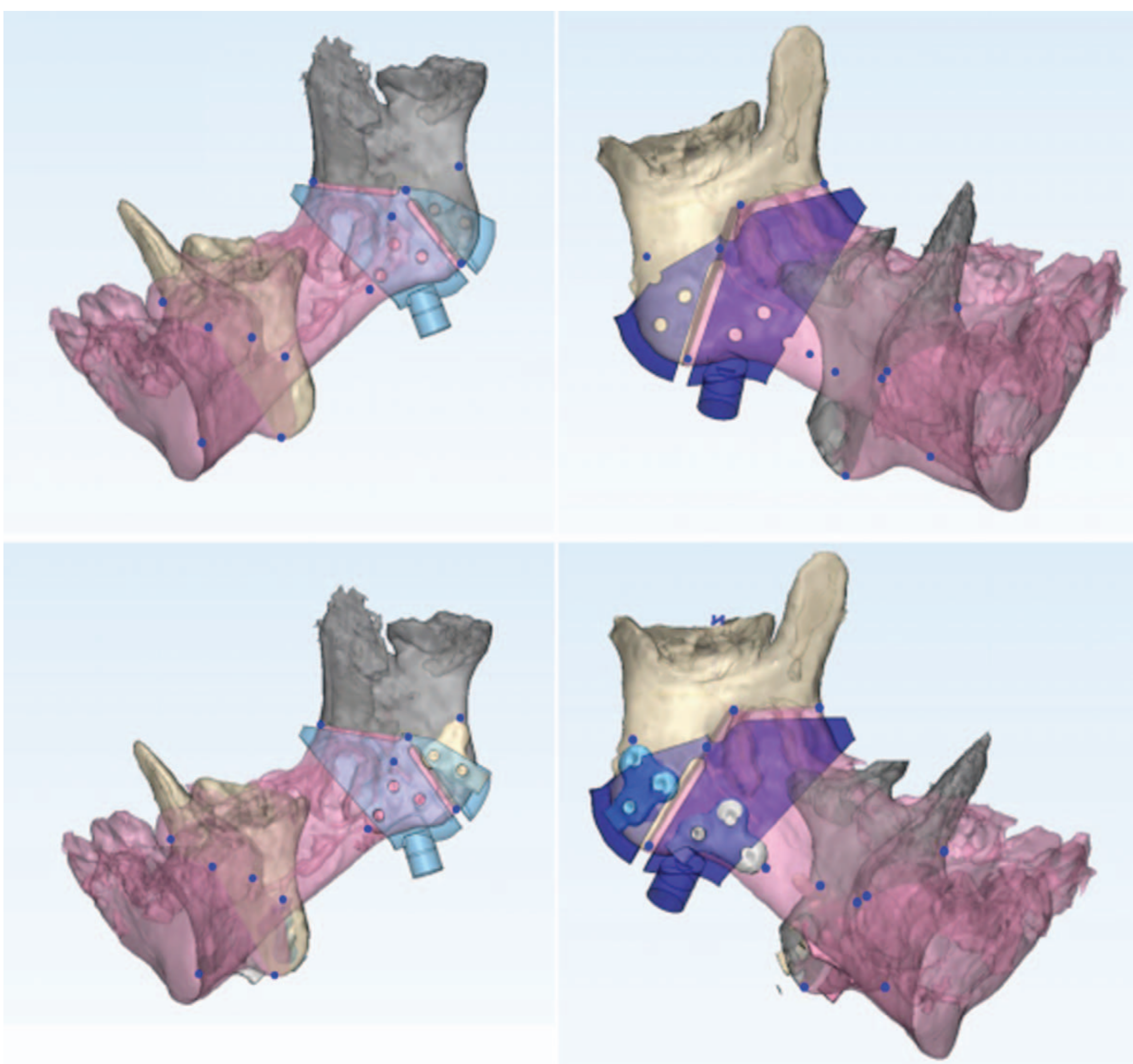

Figure 3. 3D remodeling using 3magic. A) Left mandible model with surgical guide. B) Left mandible model with surgical guide showing fixation screw positions. C) Right mandible model with surgical guide. D) left mandible model with surgical guide showing fixation screws.

Figure 4. Bilateral mandibular distraction osteogenesis using a 3D printed surgical guide. A) The incision position is marked on the patient's face. B) Morphology of the 3D printed surgical guide before use. C) The surgical guide is placed temporarily inside the incision. D) The fixation screw positions are marked by holes on the surgical guide. E) The fixation screws are placed accordingly. F) The surgical guide is removed at the end of the operation.

print group than the control group ($P=.023$), while bleeding amount (mL), time of post-surgery separation of artificial airway (days), time of hospitalization (days), and time of post-surgery oral feeding (days) did not differ ($P>.05$). Time to post-surgical separation of the artificial airway and time of hospitalization showed a trend towards a shorter time period in the experimental group ($P=.056$).

Postoperative complications were assessed by 7 parameters, local skin infection, nerve injury, tractor shedding, dental injury, accidental fracture, facial asymmetric deformity, and scarring (Table 3), none of which differed significantly between groups. However, age at operation was significantly younger in the 3D print group and surgeries were more complex, requiring a higher degree of surgical accuracy.

4. Discussion

Mandibular distraction osteogenesis was first described by McCarthy in 1989 and has since become the most widely used and effective treatment for airway obstruction caused by PRS.[14] The aim is to lengthen the mandible, which in turn will move the tongue root forward, thereby relieving respiratory tract obstruction induced by glossocoma. Successful application has been demonstrated to improve respiratory function, sleep, and swallowing.[15] It is generally performed when body position change and other conservative treatments cannot improve respiration or help wean patients from artificial respiration.[16] However, mandibular distraction osteogenesis is a high-risk treatment when used in infancy as multiple postoperative complications may occur, such as permanent tooth injury, tractor fall off, fracture accidents, mandibular alveolar nerve injury, bone nonunion, infection, and scars[17] Moreover, the severity of these congenital malformations varies widely among patients, so pre-operational individualized planning is essential to improve surgery efficacy and reduce postoperative complications. Developments in computerized structural modeling have provided surgeons with a platform to facilitate individual surgical design, but application remains difficult during actual surgery.[18] In infants, the small size of the mandible necessitates highly precise operative design and procedures, and any minor error can lead to serious complications. The use of surgical guides helps greatly in surgery design.[19] However, traditional surgical guides fabricated manually often have poor accuracy. The development of 3D printing technology may help solve this problem. By 3D printing, the surgical guide is a reverse engineered product of the surgery design. In this study, 2 different manufacturing methods of surgical guides were compared. We found that 3D printing

Table 2

Indices of surgical efficacy and safety.

Index	Control (n=12)	3D print (n=10)	*P* value
Surgery time, min	160.6667±38.1345	125.400±26.8212	.023, S
Bleeding, mL	7.5000±2.6458	8.1000±2.4698	.591, NS
Separation of artificial airway, days	5.6667±0.9847	4.7000±1.2517	.056, NS
Hospitalization, days	13.7500±1.5448	12.7000±0.8233	.068, NS
Post-surgery oral feeding, day	10.5833±1.2401	10.2000±1.3166	.491, NS

NS=no significant difference ($P>.05$), S=significant difference ($P<.05$).

Table 3

Postoperative complications.

Postoperative complications	Control (n=12)	3D print (n=10)	*P* value
Local skin infection	25%	10%	.388, NS
Nerve injury	8.33%	0%	.374, NS
Tractor shedding	0%	0%	NS
Dental injury	25%	20%	.793, NS
Accidental fracture	25%	10%	.793, NS
Facial asymmetric deformity	16.67%	20%	.849, NS
Scarring	25%	30%	.805, NS

NS=no significant difference ($P>.05$).

guide plate has obvious advantages, but the sample size of this study was small, and further enlargement of sample size is needed for comparison in the future.

Frequently used 3D printing techniques include fused deposition modeling (FDM), stereo lithography apparatus (SLA), selected laser sintering (SLS), and 3D spray (3DP). A wide range of materials can be used for 3D printing, including natural and synthesized polymers, metals, and ceramics.[20] Several processing methods can also be chosen for surgical guide synthesis, but the mechanical and biological properties will vary.[21] Choosing the optimal processing method and material composition involves many considerations. In this study, a bio-resin was used for 3D printing of surgical guides, and results demonstrated significant advantages compared to surgical guides made by traditional methods. The limitation of the investigation is the small sample size. In future studies, the sample size needs to be further expanded. So that whether this 3D printing method using bio-resin is sufficient for other clinical requirements requires additional evidence. Requirements of physical strength and dimensional accuracy vary according to operation site and surgical method. Further, the additional cost of fabrication must be balanced against clinical advantages. For different operation sites and techniques, a variety of raw materials and printing methods can be adopted for 3D printed surgical guides to lower synthesis costs while still meeting clinical requirements.

5. Conclusions

Three-dimensional printed surgical guides have sufficient accuracy and physical strength to meet the clinical requirements of difficult mandibular distraction osteogenesis. With the help of 3D printed surgical guides, we were able to shorten operative time significantly. Prospective studies are required to confirm our findings. We hypothesize that choice of the appropriate 3D printing technique and materials can reduce synthesis costs while still fulfilling clinical requirements.

Author contributions

Conceptualization: Na Zhang.
Data curation: Zhe Mao.
Formal analysis: Zhe Mao.
Project administration: Yingqiu Cui.
Resources: Na Zhang.
Supervision: Yingqiu Cui.
Writing – original draft: Zhe Mao.
Writing – review & editing: Yingqiu Cui.

References

[1] Robin P. La chute de la base de la lanque consideree comme une nouvelle cause de gene dans la respiraration naso-pharyngienne. Bull Acad Med 1923;89:37–41.
[2] Bütow K-W, Hoogendijk CF, Zwahlen RA. Pierre Robin sequence: appearances and 25 years of experience with an innovative treatment protocol. J Pediatr Surg 2009;44:2112–8.
[3] Murage KP, Tholpady SS, Friel M, et al. Outcomes analysis of mandibular distraction osteogenesis for the treatment of Pierre Robin sequence. Plast Reconstr Surg 2013;132:419–21.
[4] Flores RL, Greathouse ST, Costa M, et al. Defining failure and its predictors in mandibular distraction for Robin sequence. J Cranio-fac Surg 2015;43:1614–9.
[5] Earley M, Butts SC. Update on mandibular distraction osteogenesis. Curr Opin Otolaryngol Head Neck Surg 2014;22:276–83.
[6] McMenamin PG, Quayle MR, McHenry CR, et al. The production of anatomical teaching resources using three-dimensional (3D) printing technology. Anat Sci Educ 2014;7:479–86.
[7] Canstein C, Cachot P, Faust A, et al. 3D MR flow analysis in realistic rapid-prototyping model systems of the thoracic aorta: comparison with in vivo data and computational fluid dynamics in identical vessel geometries. Magn Reson Med 2008;59:535–46.
[8] Benum P, Aamodt A, Nordsletten L. Customised femoral stems in osteopetrosis and the development of a guiding system for the preparation of an intramedullary cavity. Bone Jt J 2010;92:1303–5.
[9] Bibb R, Eggbeer D, Williams R. Rapid manufacture of removable partial denture frameworks. Rapid Prototyping J 2006;12:95–9.
[10] Levine JP, Patel A, Saadeh PB, et al. Computer-aided design and manufacturing in craniomaxillofacial surgery: the new state of the art. J Craniofac Surg 2012;23:288–93.
[11] Ozbolat IT, Yu Y. Bioprinting toward organ fabrication: challenges and future trends. IEEE Trans Biomed Eng 2013;60:691–9.
[12] Dai X, Liu L, Ouyang J, et al. Coaxial 3D bioprinting of self-assembled multicellular heterogeneous tumor fibers. Sci Rep 2017;7:1457.
[13] Takeyasu Y, Oka K, Miyake J, et al. Preoperative, computer simulation-based, three-dimensional corrective osteotomy for cubitus varus deformity with use of a custom-designed surgical device. JBJS 2013;95:e173.
[14] Breik O, Tivey D, Umapathysivam K, et al. Mandibular distraction osteogenesis for the management of upper airway obstruction in children with micrognathia: a systematic review. Int J Oral Maxillofac Surg 2016;45:769–82.
[15] Côte A, Fanous A, Almajed A, et al. Pierre Robin sequence: review of diagnostic and treatment challenges. Int J Pediatr Otorhinolaryngol 2015;79:451–64.
[16] Viezel-Mathieu A, Safran T, Gilardino MS. A systematic review of the effectiveness of tongue lip adhesion in improving airway obstruction in children with Pierre Robin sequence. J Craniofac Surg 2016;27:1453–6.
[17] Paes EC, van der Molen ABM, Muradin MS, et al. A systematic review on the outcome of mandibular distraction osteogenesis in infants suffering Robin sequence. Clin Oral Investig 2013;17:1807–20.
[18] Mazzoni S, Marchetti C, Sgarzani R, et al. Prosthetically guided maxillofacial surgery: evaluation of the accuracy of a surgical guide and custom-made bone plate in oncology patients after mandibular reconstruction. Plast Reconstr Surg 2013;131:1376–85.
[19] Omori S, Murase T, Kataoka T, et al. Three-dimensional corrective osteotomy using a patient-specific osteotomy guide and bone plate based on a computer simulation system: accuracy analysis in a cadaver study. Int J Med Robot Comp Assisted Surg 2014;10:196–202.
[20] Tricot M, Duy KT, Docquier P-L. 3D-corrective osteotomy using surgical guides for posttraumatic distal humeral deformity. Acta Orthop Belg 2012;78:538–42.
[21] Fu J, Guo Z, Wang Z, et al. Use of four kinds of three-dimensional printing guide plate in bone tumor resection and reconstruction operation. Chin J Repar Reconstr Surg 2014;28:304–8.