

Three-dimensional biomodel use in the surgical management of basilar invagination with congenital cervical scoliosis; correction by unilateral C1-C2 facet distraction

ABSTRACT

Biomodels are produced using three-dimensional printers and their use in complex spine surgeries can be quite helpful, especially when complex anatomy is faced. In this case report, we presented a 14-year-old patient who had rigid congenital cervical scoliosis and basilar invagination and abnormalities on a neurological examination. This patient underwent atlantoaxial facet distraction and C1 C2 fusion while using a biomodel of his craniocervical junction in pre-operative planning and also as an anatomical reference per-operatively. Using biomodel in this case helped in achieving favorable surgical outcomes without any perioperative complications. Postoperative assessments including coronal deformity, basilar invagination, and neurological examination showed significant improvements and we recommend using biomodels in complex atlantoaxial distraction procedure to achieve favorable surgical outcomes with minimum complications.

Keywords: Basilar invagination, biomodel, facet distraction, scoliosis

INTRODUCTION

The biomodel is a three-dimensional representation of an anatomical structure, obtained by a technique called stereolithography (STL).^[1] The objective of this biomodel is to represent the morphology of a biological structure based on imaging studies including computed tomography (CT), magnetic resonance imaging (MRI), and then, process it in a computerized manner to obtain an anatomical prototype.^[2,3] One of the main benefits is to visualize the anatomical structures from different angles. In addition, it has proved to be a valuable tool for educating patients about their condition during the preoperative phase, also generating tactile memory for the surgeon and enabling a prior training of the technique to be used during the surgery. During surgery, it serves as a stereotactic method and anatomical reference also.^[4-7]

In this case report, we present the case of a patient with congenital cervical deformity associated with neurological symptoms, where due to the anatomical complexity of the

case, we decided to opt for a biomodel to help us with the planning and execution of the surgery.

This case is incredibly unique in its nature because it was presented with different uncommon pathologies including basilar invagination, congenital cervical scoliosis with hemivertebra, Klippel-Feil syndrome and high riding vertebral artery.

**LUIS EDUARDO CARELLI TEXEIRA DA SILVA,
DIEGO JOSÉ CUÉLLAR,
ALDERICO GIRÃO CAMPOS DE BARROS, AHSAN ALI KHAN**

Department of Spine Surgery, National Institute of Traumatology and Orthopaedics, Rio de Janeiro, Brazil

Address for correspondence: Dr. Ahsan Ali Khan,
Department of Spine Surgery, National Institute of Traumatology and Orthopaedics, Rio de Janeiro, Brazil.
E-mail: dr.ahsankhan@hotmail.com

Submitted: 05-Aug-20
Published: 26-Nov-20

Accepted: 18-Aug-20

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Teixeira da Silva LE, Cuéllar DJ, Campos de Barros AG, Khan AA. Three-dimensional biomodel use in the surgical management of basilar invagination with congenital cervical scoliosis; correction by unilateral C1-C2 facet distraction. J Craniovert Jun Spine 2020;11:347-50.

Access this article online	
Website: www.jcvjs.com	Quick Response Code 
DOI: 10.4103/jcvjs.JCVJS_131_20	

CASE REPORT

Description of the case

In this case report, after institutional review board approval (CAAE: 21225019.0.0000.5273), we described the case of a 14-year-old male patient, who consulted due to a deformity of the cervical spine and gait abnormality. On a physical examination, rigid shortening of the neck with lateralization to the left was evident. To clinically quantify the coronal deformity, we measured the head tilt (HT) using the interpupillary angle (IPA) and the shoulder tilt angle (ShTA),^[8] which were 14° and 6° respectively (both pathological since the normal value should be ≤1). On the sagittal plane, to assess total flexion deformity of the spine and its effect on horizontal gaze, we examined the chin-brow vertical angle (CBVA),^[8] which was -16° (Normal value = 1) [Figure 1]. Upper and lower limbs had normal strength on examination while the right side of the body showed hypoesthesia. Signs of upper motor neuron lesion were also observed, such as Hoffman's in upper limbs, Babinski's in lower limbs and clonus on the right side.

On radiological studies, plain radiographs and CT showed the presence of congenital scoliosis of the cervicothoracic spine, associated with Klippel-Feil syndrome and basilar invagination. Radiological measurements at cranio-cervical junction showed that the odontoid process is above the McRae, Chamberlain (3.7 mm) and McGregor (5.9 mm) lines.^[9-11] In MRI, spinal cord compression was observed [Figure 2]. All measurements of coronal balance and basilar invagination were made using Surgimap software (Nemaris Inc., New York, USA).

Given the complexity of the case, we decided to perform unilateral atlantoaxial facet distraction and C1-C2 fusion. For better preoperative planning and to have a support tool during surgery, it was decided to make a biomodel of the cervical column of the patient. This case report discusses the positive impacts of biomodel in this complex surgical procedure.

Development of the biomodel

The images for the biomodel preparation were obtained from a CT of the patient's cranio-cervical region. CT angiography was also necessary for an adequate visualization of the vertebral arteries, given their anomalous trajectory. The first stage in the construction of the biomodel was the segmentation, in which the anatomical structures were separated to facilitate the study and subsequent printing. Afterward, the DICOM file of CT was processed in the Mimics 20 software (Materialise, Leuven, Belgium), where the anatomical structures were converted to the STL format and finally sent for three-dimensional printing [Figure 3].



Figure 1: Rigid cervical deformity lateralized to the left. Interpupillary angle of 14° and shoulder tilt angle of 6° (a) Patient's dorsal view (b) chin brow vertical angle of 16° (c) Patient's lateral view (d)

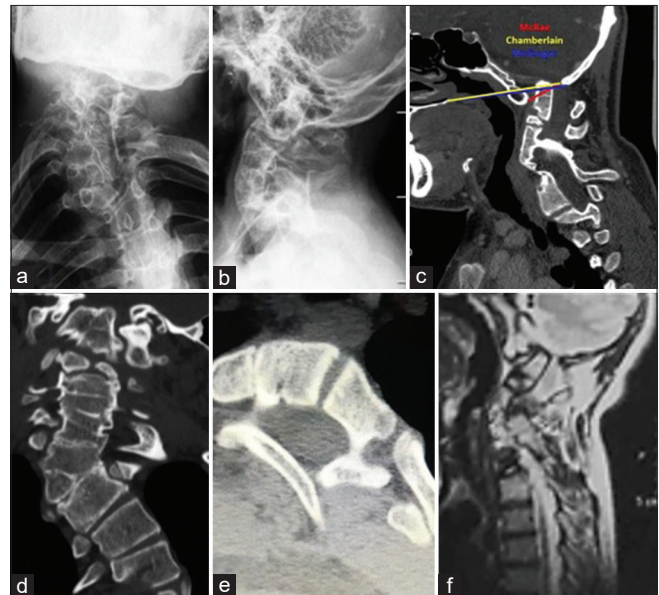


Figure 2: Anteroposterior and lateral views of cervical spine showing the scoliotic deformity (a and b) Sagittal section of the computed tomography showing protrusion of the odontoid over the lines of McRae, Chamberlain and McGregor (c) Computed tomography images showing congenital alteration in the coronal and axial plane (d and e) Sagittal magnetic resonance imaging with the signs of spinal cord compression (f)

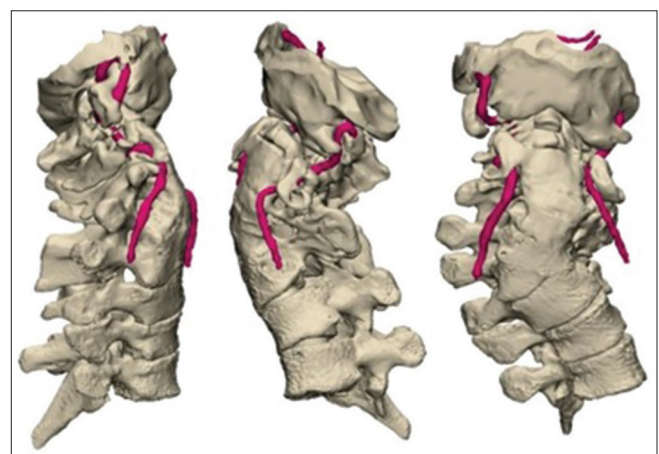


Figure 3: Computed three-dimensional reconstruction of the cranio-cervical region of the patient with the trajectories of vertebral arteries

The printing was made on a connex 500 printer using a high definition configuration (14 µm layer precision) with rigid

resin-based materials (pure white) for the base of the skull and cervical vertebrae, while a flexible material (tango) was used for the vertebral arteries. This biomodel was sterilized by sterrad sterilization system. Sterilization by this system is suited for sterilization of heat and moisture sensitive instruments since process temperatures do not exceed 50°C (140°F) and sterilization occurs in a low moisture environment. This process of sterilization takes around 1 h.^[12]

Surgical procedure

The patient was positioned in reverse Trendelenburg position with cranial traction (1/8 of body weight). A posterior approach was performed with subperiosteal exposure of the cranio-cervical junction. The C2 ganglion was identified and sectioned bilaterally to allow wide exposure of the atlantoaxial joint. The joint capsule is then opened. On the left side, joint-jamming was achieved using osteotomes and then turning 90° inside, in order to introduce a PEEK cage of 5 mm × 11 mm × 14 mm with autologous bone graft.^[13-15] During these steps, the biomodel was very useful to identify the anatomical structures [Figure 4]. On this same (left) side, we placed a screw in the lateral mass of the atlas and another one translaminar with Wright technique in C2.^[16,17]

The reason for choosing wright technique (translaminar) and avoiding pedicle was thin sized isthmus of C2 and high riding vertebral artery on the left side, and the biomodel was particularly useful during screw placement in C2, using the lamina instead of the C2 pedicle. On the right side, it was decided to perform an arthrodesis with an autologous bone graft, given the dysplasia of the C1-C2 facet joint. Due to the dangerous trajectory of vertebral artery on the right side, no instrumentation was attempted. Unilateral (left) atlantoaxial facet distraction promoted indirect spinal cord decompression by correcting HT and correction of basilar invagination with cage also corrected coronal deformity. Finally, we performed closure in anatomical planes without leaving drainage. The patient used a rigid cervical collar postoperatively for about 4 months.

Postoperative phase

Postoperatively (26 months follow-up), the patient recovered neurologically, preserved motor strength and improved



Figure 4: In addition to the physical biomodel, three-dimensional and real-time visualization of patient's reconstructed spine was available through an application of smart phones. In this figure, trajectory of vertebral artery is being identified per-operatively

gait. Signs of upper motor neuron lesion also disappeared. Clinically, a significant improvement was evident in the coronal balance of the patient, which reflected in decreased values of the IPA to 10° and the SHTA to 3°. Surgery improved the sagittal balance, reduced CBVA to -6° and improved the horizontal gaze as well [Figure 5]. Using CT scan, we reassessed the basilar invagination. We could confirm that the odontoid did not project above the cranio-cervical parameters anymore and the lines of Chamberlain and McGregor were above the odontoid (2.9 mm and 1.9 mm respectively). Postoperative images are shown in Figure 5.

DISCUSSION

The advantages of biomodels described in the surgical field have been numerous, including preoperative planning, improvement in surgical precision, reduction of complications, and an alternative to navigation during the surgery.^[18,19] A summary of the advantages reported by Martelli *et al.*^[6] in a revision of 158 studies on biomodels is shown below [Table 1].

Through our biomodel, we were able to visualize the complex osseous anomalies and abnormal course of the vertebral artery in the preoperative period and during surgery.

Table 1: Advantages of use of Biomodels

Indications	Benefits
Pre-operative planning	Direct visualization of anomalies Anticipation of per-operative challenges
Duration of intervention	Decreased operating time
Risk and complications	Less perioperative complications (blood loss, infections) Decreased radiological exposure to the patient and medical team
Outcomes	Better post-surgical results

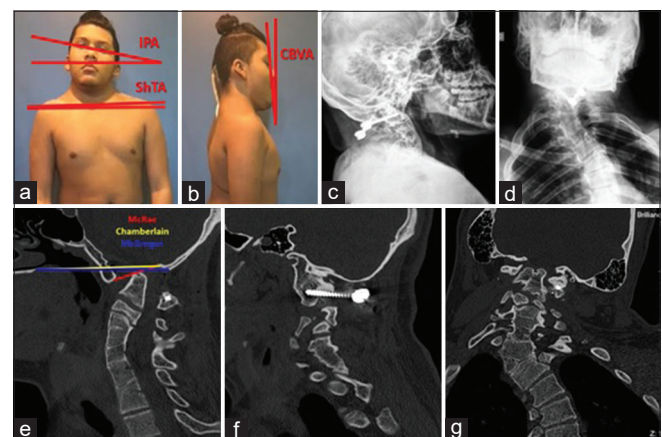


Figure 5: Clinical improvement of the coronal and sagittal balance by measuring the interpopillary angle, shoulder tilt angle and chin-brow vertical angle (a and b) Anteroposterior and lateral radiographs showing C1-C2 fusion (c and d) Computed tomography assessment of basilar invagination (e) and atlantoaxial arthrodesis (f and g)

This guide was very useful during screw placement and joint-jamming, decreasing surgery duration and reducing the blood loss (only 100 ml of blood loss). One of the limitations of using biomodel is the time required in preparation of these biomodels. Although the printing time (3–7 h) is relatively fast, the process of digital reconstruction of the anatomical structures requires more time and this processing time creates a challenge if a model is needed urgently. We believe that biomodel can be used as a guide for complex spine surgeries in limited resources where neuronavigation is not available. Using biomodel in such circumstances would not only help in planning the procedure but will also guide correct maneuvers with reducing the risk of iatrogenic complications.

CONCLUSIONS

Biomodels can facilitate preoperative planning and per-operative use as an anatomical reference in complex spine surgeries such as atlantoaxial distraction. However, integrating the use of biomodels in routine practise of spine surgery will require further research and studies with significant data.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Hull CW. Apparatus for production of three-dimensional objects by stereolithography. United States Patent, Appl., No. 638905, Filed. 1984.
- Mankovich NJ, Cheeseman AM, Stoker NG. The display of three-dimensional anatomy with stereolithographic models. *J Digital Imaging* 1990;3:200.
- Paiva WS, Amorim R, Bezerra DAF, Masini M. Application of the stereolithography technique in complex spine surgery. *Arquivos de neuro-psiquiatria* 2007;65:443-5.
- D'Urso PS, Askin G, Earwaker JS, Merry GS, Thompson RG, Barker TM, *et al.* Spinal biomodeling. *Spine* 1999;24:1247-51.
- D'Urso PS, Williamson OD, Thompson RG. Biomodeling as an aid to spinal instrumentation. *Spine* 2005;30:2841-5.
- Martelli N, Serrano C, van den Brink H, Pineau J, Prognon P, Borget I, *et al.* Advantages and disadvantages of 3-dimensional printing in surgery: A systematic review. *Surg* 2016;159:1485-500.
- Song ZL, Feng CK, Chiu FY, Liu CL. The clinical significance of rapid prototyping technique in complex spinal deformity surgery—Case sharing and literature review. *Formosan J Musculoskeletal Disorders* 2013;4:88-93.
- Kuntz C, Levin LS, Ondra SL, Shaffrey CI, Morgan CJ. Neutral upright sagittal spinal alignment from the occiput to the pelvis in asymptomatic adults: A review and resynthesis of the literature. *J Neurosurg: Spine* 2007;6:104-12.
- McRae DL. Bony abnormalities in the region of the foramen magnum: Correlation of the anatomic and neurologic findings. *Acta Radiologica* 1953;40:335-54.
- Chamberlain WE. Basilar impression (platybasia): A bizarre developmental anomaly of the occipital bone and upper cervical spine with striking and misleading neurologic manifestations. *Yale J Biology and Med* 1939;11:487.
- McGregor M. The significance of certain measurements of the skull in the diagnosis of basilar impression. *The British J Radiology* 1948;21:171-81.
- Diab-Elschahawi M, Blacky A, Bachhofner N, Koller W. Challenging the Sterrad 100NX sterilizer with different carrier materials and wrappings under experimental “clean” and “dirty” conditions. *American J Infection Control* 2010;38:806-10.
- Goel A. Treatment of basilar invagination by atlantoaxial joint distraction and direct lateral mass fixation. *J Neurosurg: Spine* 2004;1:281-6.
- Goel A, Laheri V. Plate and screw fixation for atlanto-axial subluxation. *Acta Neurochirurgica* 1994;129:47-53.
- Silva LE, Barros AG, Castro CJ, Souza RT, Azevedo GB, Casado PL. Results of the use of peek cages in the treatment of basilar invagination by Goel technique. *Coluna/Columna* 2016;15:61-4.
- Harms J, Melcher RP. Posterior C1–C2 fusion with polyaxial screw and rod fixation. *Spine* 2001;26:2467-71.
- Wright NM. Posterior C2 fixation using bilateral, crossing C2 laminar screws: Case series and technical note. *Clin Spine Surg* 2004;17:158-62.
- Malik HH, Darwood AR, Shaunak S, Kulatilake P, Abdulrahman A, Mulki O, Baskaradas A. Three-dimensional printing in surgery: A review of current surgical applications. *J Surg Res* 2015;199:512-22.
- Goel A, Jankharia B, Shah A, Sathe P. Three-dimensional models: An emerging investigational revolution for craniocervical junction surgery. *J Neurosurg: Spine* 2016;25:740-4.