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Original Article

Prediction of the functional and radiological outcome on the basis of independent factors with special emphasis on the use of 3D printed models in craniovertebral junction surgery

Rashim Kataria[1], Mudit Mehrotra[1], Devendra Kumar Purohit[1], Ajay Gupta[2], Monika Rathore[2]

Departments of [1]Neurosurgery, [2]Preventive and Social Medicine, Sawai Man Singh Medical College, Jaipur, Rajasthan, India.

E-mail: Rashim Kataria - rashim_kat@yahoo.com; *Mudit Mehrotra - moody.116@gmail.com; Devendra Kumar Purohit - devendrapurohit@rediffmail.com; Ajay Gupta - ajaykrishna193@gmail.com; Monika Rathore - rathoremonika@rediffmail.com

***Corresponding author:**
Mudit Mehrotra,
Department of Neurosurgery,
Sawai Man Singh Medical College, Jaipur, Rajasthan, India.

moody.116@gmail.com

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ABSTRACT

Background: The aim of the study was to evaluate the advantage of performing planned surgery using customized three-dimensional (3D) printed models versus performing surgery without using 3D printed models in patients with craniovertebral junction (CVJ) anomalies and traumatic CVJ fractures and dislocations.

Methods: Forty-two patients with CVJ anomalies, who were planned for operative intervention in the Department of Neurosurgery at SMS Hospital from March 2019 to February 2021, were randomly divided into two groups and analyzed. First group was operated after rehearsal on a customized 3D printed model whereas the second group underwent operative intervention without the rehearsal of surgery on the 3D printed model.

Results: Forty-two patients were enrolled for the study. Twenty-five of these patients had developmental CVJ anomalies, 16 had post traumatic Atlantoaxial dislocation (AAD), and one had congenital AAD. Twenty-three patients underwent surgical intervention using 3D printed models and 19 without using 3D printed models. The outcome in the two groups was compared using modified Japanese orthopedic association score (mJOA), recovery rate, incidence of complications such as screw malposition, postoperative neurological deterioration, vertebral artery (VA) injury, and radiological improvement based on Atlanto-Dental interval, the distance of the tip of dens from Wackhenheims clivus canal line, and the distance of tip of dens from the Chamberlain's line. The improvement in mJOA score postoperatively was found to be statistically significant in study group ($P < 0.001$) as compared to control group ($P = 0.06$). Recovery rate was better in study group than in control group ($P = 0.023$). In study group, the incidence of screw malposition and VA injury was lower than control group. Three patients deteriorated neurologically postoperatively in the control group and none in the study group. The average improvements in the radiological parameters were found to be better in study group as compared to control group postoperatively.

Conclusion: The authors conclude that 3D printed models are extremely helpful in analyzing joints and VA anatomy preoperatively and are helpful in unmasking any abnormal bony and vascular anatomy effectively, making the surgeon confident about the placement of the screws intraoperatively. These 3D models help in intraoperative error minimization with better neurological outcomes in postoperative period. In our opinion, these models should be included as a basic investigation tool in patients of CVJ abnormalities. The models also offer other advantages such as preoperative simulation, teaching modules, and patient education.

Keywords: 3D printed model, Atlantoaxial dislocation, Basilar invagination, Craniovertebral junction abnormality, Occiput–C2; C1–C2

INTRODUCTION

Craniovertebral junction (CVJ) is one of the most complex regions of the body to operate because of the limited space available for operation and unforgiving structures in the surrounding (e.g., the brain stem and vertebral artery [VA]). The course of the VA is often anomalous especially in congenital CVJ anomalies. The patient to patient variability in the size, angulation and orientation in the joints, facets and pedicle of the axis, and Atlas vertebra also adds to the complexity of this region. The use of sharp instruments and screws as well as high speed drills in this space-constrained environment makes the surgery difficult especially for a naive surgeon in this region. Surgery may lead to complications such as such as limb paralysis and VA injury leading to catastrophic hemorrhage causing life-threatening emergencies. The anatomy is further complicated by the presence of congenital osseous anomalies in this region. The risk of VA injury during CVJ surgery can be as high as 4.1%.[35] The treatment of CVJ abnormalities is aimed at correcting malalignment of bilateral C1-C2 joints in all planes, followed by stabilization of these joints.[21,26,31,32,41] Our study aims at providing evidence that surgery aided by customized 3D printed models of this region has better outcome than surgeries performed without the rehearsal of surgery on the 3D printed model.

MATERIALS AND METHODS

This prospective study was sanctioned by the ethical committee of Sawai Man Singh Medical College, and signed informed consent was taken from all patients. Forty-two patients (31 males and 11 females; mean age 24.39 ± 12.9 years for 23 cases and 28.11 ± 16.6 for 19 controls, ranging from 7 to 65 years; symptom duration 1 month – 5.5 years) with CVJ abnormalities who were admitted to our neurosurgery department between March 2019 and February 2021 were registered for the study. Clinical manifestations included neck pain (38 cases), weakness or incomplete paralysis (36 cases), posterior column involvement (30 cases), restricted neck movements (25 cases), bladder and bowel involvement (11 cases), dysphagia with dyspnea (six cases), and abnormalities in pain and temperature sensation (four cases).

Inclusion criteria

Patients presenting in outpatient neurosurgery department with CVJ anomalies planned for surgical intervention were included in the study.

Exclusion criteria

Patients presenting in emergency (those who required emergency surgeries) were excluded from the study.

Method and Materials

All patients underwent digital X-rays and computed tomography (CT) of the CVJ, CT angiography of the neck vessels, and magnetic resonance imaging (MRI) of the CVJ and cervical spine. 3D-assisted models of the CVJ along with VA were developed for 23 patients in the study groups and the remaining 19 patients were kept in control group, who underwent surgery with taking assistance from 3D models. The group allocation was done on random basis. The scans are obtained from a level extending from the occipital region up to the upper dorsal vertebra. 60 ml of iodinated contrast is injected IV at a rate of 4.5 ml/s. Axial 0.8-mm-thickCT images are obtained at 0.5-mm intervals. These data are in DICOM format. The angiography images of these patients are used to extract 3D file in Surface Tessellation Language (STL) format. This STL file was then sent electronically to a local laboratory with a 3D printer station for printing of 3D model [Figure 1]. The models are prepared using Acrylonitrile Butadiene Styrene (ABS) polymer by a fused dependent modeling (FDM 3D) printer. The FDM 3D printer used was ProtoCentre999.

The preoperative and the postoperative values of the craniometric indices: Atlantodental interval (ADI), the distance of the tip of dens from Wackhenheims clivus canal line (WCCL), and finally the distance of tip of dens from the Chamberlain's line (CL) were calculated in both the

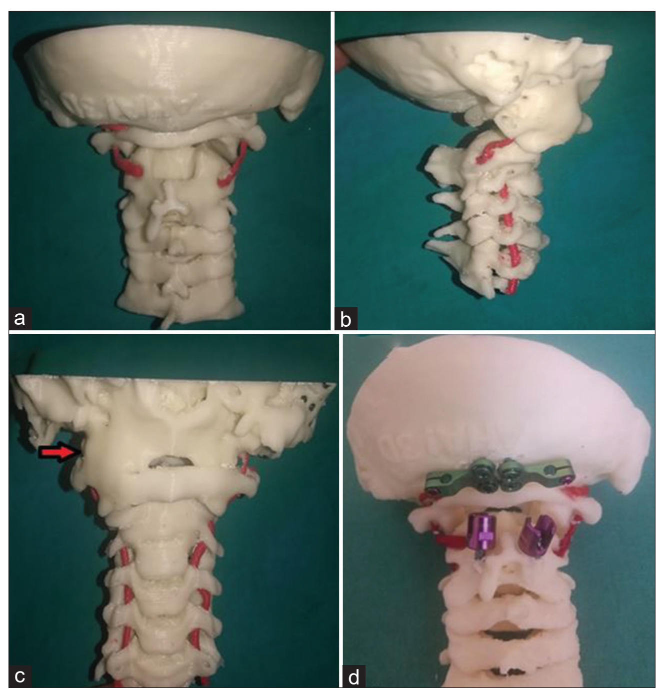

Figure 1: (a) Posterior view of a three-dimensional printed model of a patient, (b) lateral view of the model, (c) anterior view of the model showing fused left lateral mass of C1 with occipital condyle (solid red arrow), and (d) practice on model with occipital-C2 plate and screws.

study and the control groups. The neurological assessment of patients in the preoperative and the postoperative period was done using the modified Japanese orthopedic association score (mJOA) using motor and sensory functions of the four extremities and the sphincter, which amounted to a total of 18 points [Figure 2].[30] Using the modified JOA score, the recovery rate[12] was calculated for every patient in each group using the formula

$$Recovery\ rate = \frac{Postoperative\ m\ JOAS - preoperative\ m\ JOAS}{Total\ m\ JOAS - preoperative\ m\ JOAS}$$

Screw malposition was an important parameter compared between the two groups. Criteria used for deciding screw malposition were based on the radiological findings in postoperative CT scans and on the neurological status of the patient. The radiological criteria were any cortical breach of pedicle on medial side, cortical breach on lateral and inferior side of more than 2 mm, and finally cortical breach superiorly and anteriorly more than 4 mm. These criteria's were established using McGill's scoring system for revision of pedicle screws.[3]

SCORE	DEFINITION
	Motor dysfunction score of the upper extremities
0	Inability to move hands
1	Inability to eat with a spoon, but able to move hands
2	Inability to buttom shirt, but able to eat with a spoon
3	Able to button shirt with great difficulty
4	Able to button shirt with slight difficulty
5	No dysfunction
	Motor dysfunction score of the lower extremities
0	Complete loss of motor and sensory function
1	Sensory preservation without ability to move legs
2	Able to move legs, but unable to walk
3	Able to walk on flat floor with a walking aid (i.e., cane or crutch)
4	Able to walk up and/or down stairs with hand rail
5	Moderate to significant lack of stability, but able to walk up and/or down stairs without hand rail
6	Mild lack of stability but walks with smooth reciprocation unaided
7	No dysfunction
	Sensory dysfunction score of the upper extremities
0	Complete loss of hand sensation
1	Severe sensory loss or pain
2	Mild sensory loss
3	No sensory loss
	Sphincter dysfuntion score
0	Inability to micturate voluntarily
1	Marked difficulty with micturition
2	Mild to moderate difficulty with micturition
3	Normal micturation

Figure 2: Modified Japanese orthopaedic association score (m JOA), the maximum score is 18.

The underlying principle of this surgery was neural decompression with stabilization of the CVJ complex. The posterior internal fixation surgery was done in each patient according to preoperative CT and MRI findings. In patients with Atlanto-occipital assimilation, internal fixation was performed using occipital plate and C2 pedicle screws, as first described by Olerud *et al.*[22] It was done in 13 patients in study group and in eight in control group. C1 lateral mass screw (inserted in 10 patients of study group and 11 patients of control group) and C2 pedicle screw fixation (inserted in 14 patients of study group and 14 patients of control group) using Goel's and Harm's technique[8,11] were the procedure of choice in patients without atlanto-occipital assimilation. Intra-articular spacer or autologous bone graft was used in patients in both the groups wherever needed. In case, the pedicles of the C2 were not of adequate size, or in cases of high riding VA, C2 translaminar screws using Wright's technique,[36] or C2 pars interarticularis screws were used (inserted in six patients of study group and three patients of control group). C3 lateral mass screw fixation (occiput-C2-C3) was added in some traumatic cases to reinforce the construct (inserted in three patients of study group and two patients of control group).

The patients were followed up for a period of 1 year. Follow-up X-ray and CT of the neck were performed to investigate fusion maturation and bone growth at 1–3 months. Fusion was defined as presence bone trabeculae between the C1 and C2 facets without the presence of any gap. Cystic lucencies around the implants or along the endplates and linear defects within the bridging trabeculae suggested nonfusion.

Statistics

Data were entered into excel sheet. Continuous data were summarized in the form of mean and standard deviation. Difference in mean of two groups was analyzed using student "*t*" test. Discrete data were expressed in the form of proportions and difference in proportions was analyzed using Chi-square test. The level of significance was kept 95% for all statistical analysis.

RESULTS

Forty-three patients (31 males and 11 females) were enrolled for the study. Twenty-five of these patients had developmental CVJ anomalies, 16 presented as posttraumatic Atlantoaxial dislocation (AAD), and one had AAD with no history of trauma or developmental deformity. Twenty-three patients underwent surgical intervention using 3D printed models and 19 without using the aid of 3D printed models. The two groups were compared using mJOA, recovery rate, incidence of complications such as screw malposition, postoperative neurological deterioration, VA injury and radiological improvement based on ADI, CL, and WCCL.

The improvement in modified JOA score postoperatively as compared to the preoperative score was found to be statistically significant in study group [Figure 3a] ($P < 0.001$) as compared to control group [Figure 3b] ($P = 0.06$) [Table 1]. The recovery rate was calculated for each patient using the formula stated above. The patients in both groups were divided into four groups according to the recovery rate, that is, ≤25%, 25–50%, 51–75%, and 76–100% and were then compared. Better improvement in recovery rates was found in the study group than in control group and this was found to be statistically significant ($P = 0.023$, i.e., <0.05) [Figure 4a and Table 2].

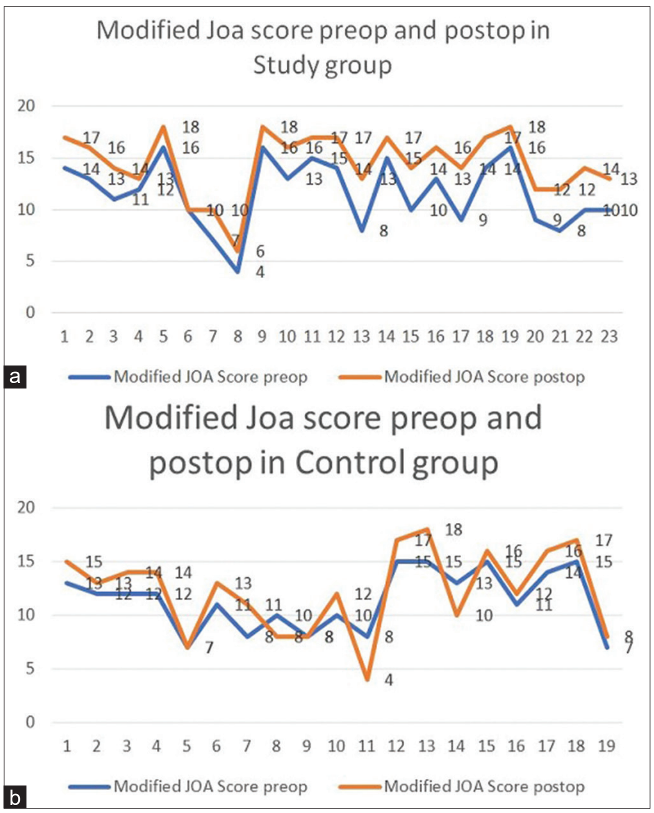

Figure 3: (a) Comparison of preoperative and postoperative m JOA score in study group and (b) Comparison of preoperative and postoperative m JOA score in control group.

The improvement in the postoperative values of the craniometric indices, such as ADI, CL, and WCCL, was compared to preoperative data. It was found to be statistically significant in both the study and the control group but then on comparing the mean of these three indices in both preoperative and postoperative period, the improvement was found to be better in the study group versus the control group; ADI (3.94 in study vs. 3.15 in control group) [Figure 4b], CL (4.2 in study vs. 2.76 in control group) [Figure 4c], and WCCL (4.55 in study vs. 3.23 in control group) [Figure 4d].

After excluding the occipital screw and plate insertion in each patient in both study and control group, screw malposition was studied in both groups. It was observed that only two screws (out of 66 total inserted screws) were malpositioned in the study group (according to the McGill's criteria), as compared to eleven screws (out of total 60 inserted screws) found to be malpositioned in the control group. This was found to be statistically significant ($P = 0.012$, i.e., <0.05) [Table 3].

In the study group, almost all of the patients improved neurologically in the postoperative period as evident by their improved mJOA scoring. There was symptomatic improvement in neck pain and neck movements. There was no incidence of VA injury. One of our cases in study group had malpositioned C2 pedicle screw medially and did not improve neurologically in postoperative period, but there was no evidence of any neurological deterioration in this patient. The patient was advised for screw revision but the patient refused surgery. One of our pediatric patients had a single episode of Generealized Tonic-ClonicSeizures in the postoperative period, his CT scan brain showed no abnormality, and the patient was discharged without any neurological deficit. None of these patients had any implant failure requiring removal of the implant, wound-related complications, or cerebrospinal fluid leakage.

In the control group, out of 19 patients, eight patients showed screw malposition (using the McGills's criteria). Four of these patients showed only minor improvement in their neurological status in postoperative period as depicted by their postoperative mJOA score and recovery rate. No improvement was seen in other two patients. Three patients

Table 1: The improvement in m JOA score in postoperative period as compared to preoperative period was statistically significant in study group (P≤0.001) and not significant in control group (P=0.06).

Variables	Study Group		*P*-value	Control Group		*P*-value
	Pre op	Post op		Pre op	Post op	
Modified JOA score	11.61±3.26	14.43±3.07	<0.001	11.37±2.81	12.26±3.93	0.06
Atlantodental Interval	8.58±2.83	4.64±2.51	<0.001	7.34±2.28	4.19±2.73	<0.001
Chamberlains Line	3.10±8.83	−1.10±5.91	0.005	1.61±7.84	−1.15±5.47	0.004
Wakenheims clivus canal line	4.15±9.24	−0.40±5.45	0.006	1.94±4.64	−1.29±4.88	0.005

The preoperative and postoperative ADI, CL, and WCCL were found statistically significant in both study and control group

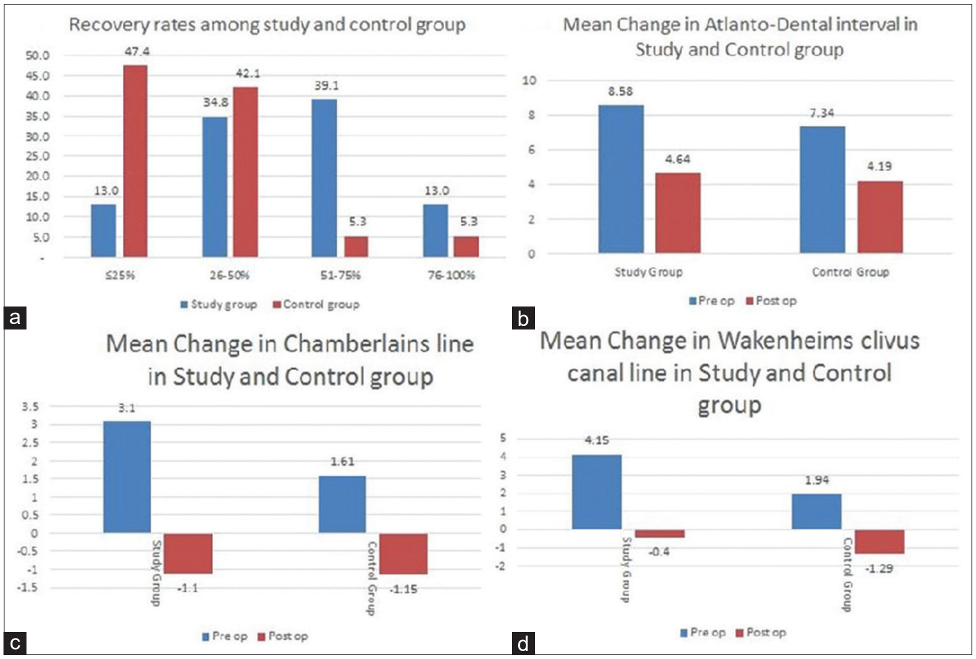

Figure 4: (a) Comparision of recovery rates among study and control group, (b) mean change in atlanto-dental interval in study (3.94) versus control group (3.15), (c) mean change in canal line in study (4.2) versus control group (2.76), and (d) mean change in wackhenheims clivus canal line in study (4.55) versus control group (3.26).

Table 2: Recovery rate postoperatively was found to be better in the study group as compared to the control group and was statistically significant (*P*=0.023).

Variable	Study group (*n*=23)		Control Group (*n*=19)		*P*-value
	Number	Percentage	Number	Percentage	
Age					
0–15	7	30.43	6	31.58	χ^2=0.227, *P*=1
16–30	10	43.48	7	36.84	
31–45	4	17.39	4	21.05	
≥46	2	8.70	2	10.53	
Gender					
Male	17	73.91	14	73.68	χ^2=0.113, *P*=0.737
Female	6	26.09	5	26.32	
History of Trauma					
Present	7	30.43	9	47.37	χ^2=0.649, *P*=0.420
Absent	16	69.57	10	52.63	
Congenital anomaly present					
Present	14	60.87	11	57.89	χ^2=0.014, *P*=0.904
Absent	9	39.13	8	42.11	
Recovery Rate					
≤25%	3	13.04	9	47.37	χ^2=10.111, *P*=0.023
26–50%	8	34.78	8	42.11	
51–75%	9	39.13	1	5.26	
76–100%	3	13.04	1	5.26	

deteriorated neurologically in the postoperative period of which one patient expired due to respiratory failure on 3rd postoperative day, his head CT was suggestive of brain stem and cerebellar infarction.

The results showed that posterior internal fixation performed using individualized 3D printed model is realistic and effective in treating CVJ abnormalities [Figure 5]. The 3D printed models have found more utility in patients with CVJ congenital abnormalities as compared to patients presenting with traumatic CVJ fractures as the later have relatively straightforward anatomy of this region. The 3D printed model is clinically advantageous and may have great potential in guiding surgery in this region, even to the relatively less experienced neurosurgeons in the field.

DISCUSSION

The occipitoatlantal joint provides more stability in comparison to mobility and the reverse is true for the atlantoaxial joint, which offers more mobility leading to higher chances of injury and dislocation at this joint.[34] The patients with C1-C2 instability (AAD) present with a plethora of symptoms ranging from neck pain and restricted neck movements to numbness and weakness in the upper and lower limbs.[20,24,25,28.39] The osseous, ligamentous, neural, and vascular anatomy in this region is complex and variable. The VA can be anomalous in 2–3% in normal population and in 20% of congenital CVJ anomalies.[5,19] Occipitalization of atlas is seen in 0.1–0.8% of normal population[18] while it is considerably higher in patients with AAD.[4,14,15] Sardhara *et al*.[27] in his study found occipitalization of atlas in 72% of his enrolled CVJ anomaly patients. It has been seen that occipitalization of atlas and C2–C3 fusion is frequently associated with lateral joint asymmetry due to their common embryological development.[27] This frequently leads to facetal instability and consequently AAD.[9] C2 bony anomalies include segmentation defects of the odontoid process such as osodontoideum and uncommonly absence of C2 posterior elements leading to C2-C3 spondyloptosis[16] [Figure 5].

Table 3: Incidence of screw malposition was very low in study group compared to control group and this was found to be statistically significant.

	Screw Malposition		*P*-value
	Present	Absent	
Study group	2	64	χ^2=6.386, *P*=0.012
Control group	11	49	

The surgical management of AAD has seen a paradigm shift from transoral decompression followed by fixation to now

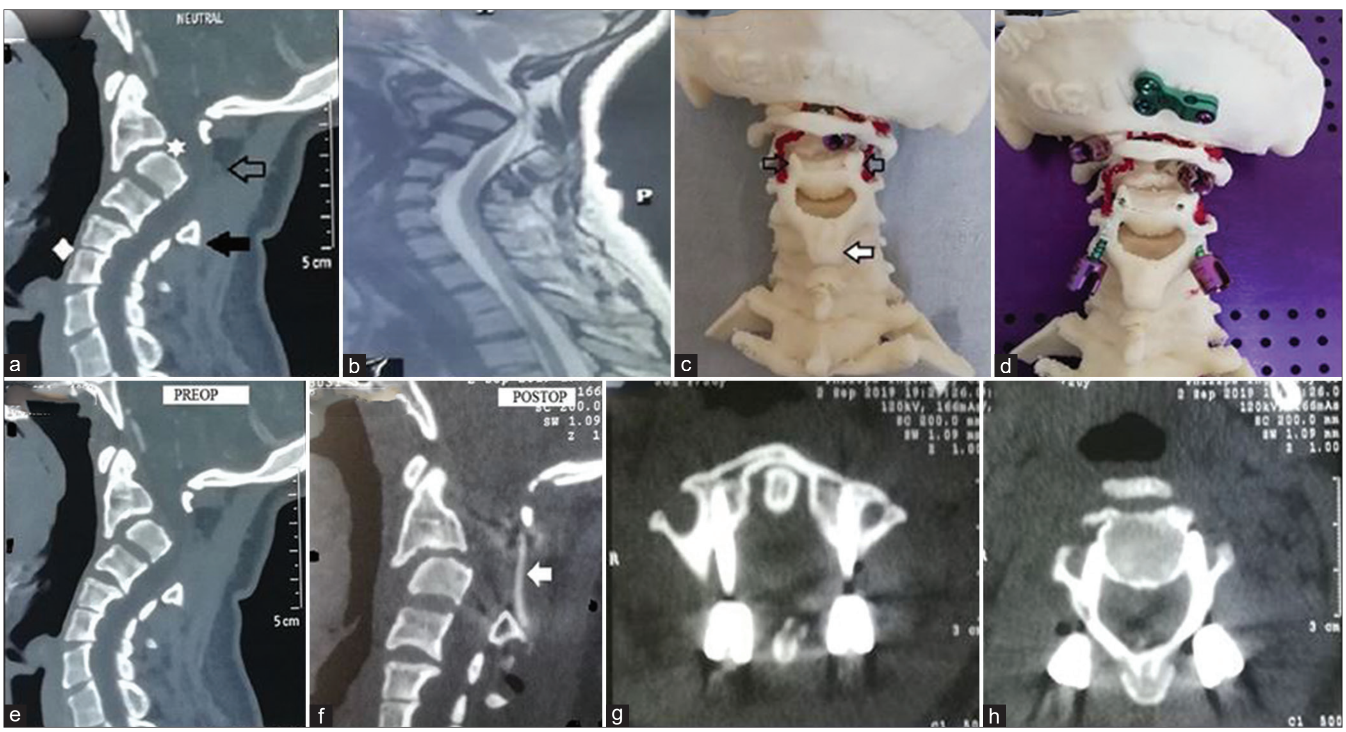

Figure 5: Uncommon case of absent C2 posterior elements with C2-C3 spondyloptosis; (a) preoperative computed tomography (CT) scan (MidSagittal cut) showing absent C2 lamina (hollow arrow), C2-C3 spondyloptosis (asterix), hypertrophied spinous process of C3 (solid arrow), and C5-C6 block vertebra (diamond), (b) magnetic resonance imaging T2W image, midsagittal section showing severe cord compression by retropulsed C3 body, (c) 3D model of the craniovertebral junction of the patient showing posterior view with absent C2 lamina, well developed uncinate process of C3 (hollow arrow), and hypertrophied spinous process of C3 (solid arrow), (d) showing hands on practise on the model preoperatively, (e and f) comparison between the preoperative and postoperative CT scan (Mid Saggital view) of the patient showing acceptable reduction and realignment of the C2-C3 body with increased canal diameter at the level of C3 and iliac bone graft between C1 posterior arch and C3 spinous process *in situ* (solid arrow). The model helped us in better understanding of the complex anatomy and planning preoperatively, and (g and h) C1 lateral mass and C2 pedicle screw insertion.

more popular posterior fixation approaches using the screw-rod system, which includes pedicle screw system; lateral mass screws system, transarticular screws system, and laminar screws system. The most commonly used method of internal fixation for posterior cervical surgery is the pedicle screw system because of its excellent biomechanical property and the ease of surgery.[27]

The 3D reconstructed CT images are better than the 2D pictures and can help in understanding the complex anatomy but they are of limited use intraoperatively as they can be viewed only on a CT film and provide with only 2D prospective of the region concerned. The newer neuronavigation devices such as CT 3D navigation system, G-arm, and O-arm are able to serve the purpose of providing a 3D perspective of the region intraoperatively but the practical applications of these devices are limited by factors such as lack of equipment, insufficient training, and high costs.[38,40] Introduction of the 3D printing technology in the field of neurosurgery has helped the surgeons in better understanding of the complex anatomy of different regions of the patients. Even though in its infancy, the 3D printing technology is proving to be of immense help in understanding the complex anatomy of the CVJ, study the course of VA, evaluate the dimensions of the bones, and their orientation for identifying the best possible trajectory of screw implantation in posterior fixation surgeries. Despite the biomechanical superiority of cervical pedicle screws,[38] the placement of cervical pedicle screws has a considerable risk of injury, either to nerves or to the VA. Abumi *et al*.[1] reported that 45 out of 669 inserted screws (6.7%) were misplaced in their early series. Onishi *et al*.[23] reported on a patient who suffered cerebral infarction due to brain embolism after the placement of cervical pedicle screws. Having a tangible model of a patient's anatomy available for a surgeon to study or use to simulate surgery is preferable to the costly and not easily available neuronavigation techniques used for assistance in surgeries in this region. The benefit was also observed in our study, where patients operated using 3D models had no neurological deterioration postoperatively. Guo *et al*.[10] found a greater acceptable rate of screw placement in 94.6% of the patients in which 3D model-based navigation templates were used as compared to a 70.27% satisfactory rate in the control group in which screws were inserted using fluoroscopy. In our study, only two screws were misplaced in study group as compared to 11 in control group. Yang *et al*.[37] in his study stated that screw positions were incorrect in 32.9% of patients who were treated with conventional free-hand techniques as compared to 11.3% of patients who were treated with screw fixation assisted by 3D-printed models. Wright and Lauryssen[35] reported that VA injury can be as high as 4.1% per patient or 2.2% per screw inserted, which can be drastically reduced using a 3D models based intraoperative navigation. Chhabra *et al*.[5] in his study concluded that these 3D printed model provides an excellent window to study VA relationship to C1–C2 joint, thereby helping the surgeon immensely during the intraoperative period. According to a study by Sugimoto *et al*.,[29] the effectiveness of 3D printing escalates many folds with the complexity of the pathology, as the surgeon is able to better appreciate the patients anatomy with the life size model in his hands as compared to reconstruct the patients anatomy in his mind during surgery using the preoperative CT scan images. Izatt *et al*.[13] in his study found that the use of a 3D printed model improved surgical results in 78% of cases, he also stated that the additional costs incurred during manufacturing of these 3D printed models were counterbalanced in 59% of cases by the more proficient operative technique and planning afforded by the models, as well as fewer intraoperative complications and higher precision associated with the use of these 3D models. This study was supported by Wang *et al*.,[33] who stated that the diminished need for intraoperative navigation reduced the operating costs of the procedure. Similar encouraging results have be shown by Goel *et al*.,[7] Gao *et al*.[6] and Kataria *et al*.[2,17] in their studies, where they have concluded that individualized 3D printed model–assisted posterior internal fixation is more effective in improving the treatment of CVJ abnormalities and help in lowering intraoperative as well as postoperative complications.

The 3D model can be used to practice the operation before the actual surgery, the size of the plates and screws to be used and the angle of insertion of the screws can be calculated preoperatively from these 3D models. The model also provides information about the overriding joints angulation, the size of the screwable structures such as pars, pedicle, lateral masses, and lamina. The entry points of the screws can be selected and the screws can be inserted in the model. This saves the operative time during the actual operation and also reduces the blood loss.[39] A Plan B can be always be decided before the actual operation, in case the primary surgical plan fails. The rehearsal minimizes the surprises encountered during the actual operation. Repeated attempts of screw insertion in the model reduce the chances of screw malposition during the operation on the patient. A naive surgeon is definitely more comfortable operating on a patient after doing the rehearsal on the same anatomy provided by the 3D model. He has a sense of familiarity with the anatomy during the actual surgery.[39-41] Last but not the least, these 3D models can also be used as teaching tools for the residents in institutions and act as aids for patient education and counseling preoperatively.[39-41]

Even though 3D printed models provide invaluable information but they have some limitations too. The most of the models are made of single material, that is, plastic (ABS), hence are brittle, they do not yield to the forces and manipulations (e.g., DCER) and break off; hence, the actual targeted correction of the deformity cannot be achieved in the model. The joints in the model are immobile and, hence, are unable to provide information with regard to the nature of the joint instability and flexibility. No information regarding the venous anatomy and the extent and size of venous plexus in the region of lateral gutters could be deciphered from the models. The model provides details about the osseous anatomy only and does not provide information on the strength and integrity of various ligaments and soft tissues. In redo cases, models with a single material are not able to distinguish between the bones and the implant.

CONCLUSION

Even though there are few shortcomings with 3D models, they provide with hands-on approach preoperatively and help immensely in decoding the complex anatomy of the CVJ which can help us in achieving a complication-free surgery in an otherwise complex region. This is even more relevant in cases of congenital CVJ anomalies where the anatomy is severely deranged and there are more chances of anomalous course of VA. The surgeon can have an excellent understanding of the anatomy by having hands on experience on the model. With further advances in the technology, it can be stated with reasonable surety that 3D printer will continue to prove itself as an important and highly cost-effective tool in the management of CVJ lesions.

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- All authors of this research paper have directly participated in the planning, execution, and analysis of this study
- All authors of this paper have read and approved the final version submitted and each author believes that the manuscript represents honest work
- No conflict of interest relevant to this article was reported
- The content of this manuscript has not been copyrighted or published previously
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- There are no directly related manuscripts published or unpublished by any authors of this paper
- My institute's (SMS Medical College) representative is fully aware of this submission
- My research project was not sponsored by any agency.

Declaration of patient consent

Patients' consent not required as patients' identities were not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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