

# Surgical treatment for upper cervical deformity with atlantoaxial joint dislocation using individualized 3D printing occipitocervical fusion instrument

# A case report and literature review

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# Abstract

To introduce a novel technique of using individualized 3D printing occipitocervical fusion instrument (3D-OCF) for the treatment of upper cervical deformity with atlantoaxial joint dislocation.

The surgery for deformity of the craniocervical junction area is a challenge in the field of spine. If the surgical deviation is too large to injure the spinal cord or vertebral artery, it will cause catastrophic damage to the patient. Therefore, it is controversial whether these patients should undergo surgical treatment. We provide a novel surgical approach for the challenging upper cervical surgery through 3D-OCF and a typical patient.

We present a 54-year-old female patient, who suffered from dizziness and numbress in her limbs for 8 months. After the patient was admitted, we performed the three-dimensional CT scan, modeled using Mimics software 17.0, and designed customized occipitocervical fusion instrument. Besides, we repeatedly perform simulated surgery based on 3D-printed models before surgery.

The operative time was 142 minutes and the intraoperative blood loss was 700 mL. X-ray showed reduction of atlantoaxial dislocation and accurate position of internal fixation. The patient's symptoms were significantly relieved: the sensation of dizziness and numbress of limbs was obviously relieved, and the sense of banding in chest, abdomen, and ankle was disappeared. At the last follow-up, imaging showed that 3D-OCF had bone-integration and Syringomyelia was disappeared. The patient's cervical JOA (Japanese Orthopaedic Association) score increased from 10 points to 17 points.

Individualized 3D-OCF can improve the safety and accuracy of upper cervical surgery, reduce the operative time and the number of fluoroscopy. Our study provides a novel surgical approach for the challenging upper cervical surgery.

Abbreviations: 3D-OCF = 3D printing occipitocervical fusion instrument, JOA = Japanese Orthopaedic Association.

Keywords: 3D printing, individualization, occipitocervical fusion, upper cervical deformity

Editor: Arjun Ballal.

All data are fully available without restriction.

The author information can be found in the title page.

Ethics and governance approval for this study was obtained from Ethics Committee of Bengbu Medical College and the Second Affiliated Hospital of Bengbu Medical College.

The authors have no conflicts of interest to disclose.

The datasets generated during and/or analyzed during the current study are publicly available.

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Received: 29 November 2020 / Received in final form: 20 February 2021 / Accepted: 25 February 2021

http://dx.doi.org/10.1097/MD.00000000025202

This project is funded by Anhui Provincial Department of Science and Technology (www.ahkjt.gov.cn), funding numbers: 1804h08020247, and the Key Laboratory of Digital Orthopaedics, Bengbu Medical College.

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How to cite this article: Niu G, Chen H, Liu L, Zhou G, Zhou Q, Li C, Dai J, Nie H, Bai J, Zhang J. Surgical treatment for upper cervical deformity with atlantoaxial joint dislocation using individualized 3D printing occipitocervical fusion instrument: a case report and literature review. Medicine 2021;100:12(e25202).

# 1. Introduction

The deformity of the craniocervical junction area leads to the variation and complexity of local anatomy. Besides, this area includes important structures such as spinal cord and vertebral arteries. Currently, it is difficult to accurately place upper cervical pedicle screws with traditional free-hand methods, even for experienced spine surgeons. When surgical deviation is too large to injure the spinal cord or vertebral artery,<sup>[1,2]</sup> it will cause catastrophic damage to the patient. Therefore, it is controversial whether these patients should undergo surgical treatment.

Although Da Vinci surgery has been invented, its equipment is expensive and not convenient to promote in general hospitals. What's more, the traditional screw-rod system requires a large number of bone grafts, and the case of broken screw-rod occurs sometimes.<sup>[3]</sup> Therefore, it is worth studying to explore a new technique of posterior occipitocervical fusion. We use modern computer imaging technology, reverse engineering technology, and 3D printing rapid prototyping technology to design a 3D-OCF based on titanium powder (Patent Number: ZL201721220260.5), shown in Figure 1. The fusion instrument combines anatomical customized titanium plate fixation, individualized guided screw placement, and bionic microhole surface fusion, which can accurately place the screw to the upper cervical pedicle.<sup>[4,5]</sup> Meanwhile, the 3D-printed porous material can promote bone fusion, improve the fusion rate, and reduce the risk of broken nails and rods.<sup>[6]</sup> In the following, we present a typical patient with upper cervical deformity to show the specific steps of the operation.

## 1.1. Patient history

A 54-year-old female patient suffered from dizziness and numbness in her limbs for 8 months due to trauma. When admitted, she complained the symptom progressed and cannot walk smoothly. Physical examination: the patient felt stiffness in neck, numbness in both hands, and severe symptoms on her right side; besides, she also felt the sense of banding in the chest, abdomen, and ankles; symptoms vary with body position, neck flexion worsens, and extension symptoms ease; Hoffman's and Babinski signs were negative bilaterally; there was no obvious restriction on the active flexion and extension of the limb joints and normal muscle tension; the preoperative JOA (Japanese Orthopaedic Association) score was 10 points.

# 1.2. Imaging

Dynamic radiographs showed congenital atlantoaxial ossification (Fig. 2 A–D). Cervical CT three-dimensional reconstruction showed that atlas and occipital foramen fusion, slightly enlarged anterior atlas space, suspected congenital atlas occipital ossification, and atlantoaxial subluxation (Fig. 2E). Cervical MRI demonstrated that the tonsil of cerebellum under the tentorium became sharp, the herniation moved down into the foramen magnum, the spinal cord was compressed, the central canal was expanded, and the area of the cavity reached 13 mm × 8 mm (Fig. 2F–H).

We give the following diagnosis based on the patient's medical history, physical and imaging examination. Clinical diagnosis: Chiari malformation, Syringomyelia, Congenital occipital ossification of atlas, and Atlantoaxial dislocation.

# 2. Materials and methods

# 2.1. Data acquisition and processing

The three-dimensional CT data of the patient's cervical spine was imported into Mimics 17.0 (Materalise Company, Belgium) for model reconstruction, and the dislocated atlantoaxial joint was simulated to reduction (Fig. 3A–D). Using virtual screws to place nails on 3D reconstructed models in Mimics 17.0 software.

# 2.2. Design of virtual digital occipitocervical fusion instrument

Reverse engineering software NX 10.0 was used to design a reverse template based on the anatomy of the atlantoaxial, the skull base, and the dorsal side of the C3 lamina. Design the optimal screw entry point and channel according to coronal, sagittal, and horizontal positions (Fig. 4A, B), and fit it with the reverse template. Making an individualized posterior occipitocervical fusion instrument with positioning and orientation holes (Fig. 4C, D). On the reconstructed cervical spine model, the fusion device was fitted to the back of the corresponding vertebral body, and the degree of matching between the positioning guide hole and the pedicle nailing point was observed from different angles (Fig. 5A–D), and the STL file was derived after the matching.



Figure 1. 3D printing occipitocervical fusion instrument.



Figure 2. A–D, Dynamic radiographs showed congenital atlantoaxial ossification. E, Atlantooccipital ossification with atlantoaxial subluxation. F–H, Upper cervical spine on MRI.

#### 2.3. Inspection

The technical specification report of the occipitocervical fusion instruments and preselected screws is sent to the surgeon, and the surgeon makes a modification according to the actual situation of the surgery. This process is reiterated until the surgeon confirms the design and provides approval for production.

## 2.4. Making customized occipitocervical fusion instrument

Using titanium powder as material, the instrument (Aikang Medical Company, Beijing, China) was printed on a medical industrial metal 3D printer. Matching and screw placement tests

were performed on the 1:1 vertebral body model (polylactice acid material). If the fusion instrument does not match the upper cervical spine or the accuracy of screw placement is poor, the fusion device will be fine-tuned and improved; the fusion device with suitable test compatibility is routinely sterilized for surgery.

# 2.5. Surgical procedures

The novel surgical approach has been reviewed by the Ethics Committee of the Second Affiliated Hospital of Bengbu Medical College, and the patient signed the consent form. The patient was placed in the prone position and the general anesthesia with



Figure 3. A-D, Modeling and reduction of atlantoaxial joint by Mimics 17.0.



Figure 4. A, B, The optimal screw entry point and channel according to coronal, sagittal, and horizontal positions. C, D, Making an individualized posterior occipitocervical fusion instrument with positioning and orientation holes.

endotracheal intubation was performed. The skull around the occipital protrusion, the spinous process, lamina, and lateral mass of C3-4 were fully exposed by incision of the posterior midline. We used an ultrasound scalpel to remove the posterior arch of the atlas, enlarge the foramen magnum, and traction the skull to restore the atlantoaxial joint. The 3D printing occipitocervical fusion instrument was utilized to test the model. We found that the surface of the occipital-neck after the reduction was completely attached to the test mold. Grind the surface of the skull, C2/3 spinous processes, lamina, and lateral mass with a drill to remove the cortex. A 1.0 mm diameter Kirschner wire was used to drill micro holes on the surface of the cranial neck, and the depth was suitable to drill through the outer plate of the skull. The 3D-printed fusion instrument was placed at the back of the occipital neck, and a guide sleeve was installed in the pedicle screw hole. This guide was used to drill holes under the C-arm lateral perspective monitoring. The C2/3 pedicles were respectively placed into 2 screws. The reduction of atlantoaxial joint and the suitable internal fixation were observed under the fluoroscopy (Fig. 6A–D). We place an incision drainage strip and suture the incision.

#### 3. Results

The operative time was 142 minutes and the intraoperative blood loss was 700 mL. The patient's symptoms were significantly relieved: the sensation of dizziness and numbness of limbs was obviously relieved, and the sense of banding in chest, abdomen, and ankle was disappeared. The radiographs of cervical vertebrae demonstrated that atlantoaxial reduction, normal physiological curvature, and occipitocervical fusion fixation system was well fixed (Fig. 7A, B). Ten days after operation, the neurological symptoms of the patients were significantly improved. CT showed that the internal fixation was good, the atlantoaxial vertebra was completely restoration, and the foramen magnum was enlarged (Fig. 7C); MRI showed that the cervical spinal cord angle of medulla oblongata returned to normal and the extent of syringomyelia was significantly smaller than that before operation (Fig. 7D). At 6 months postoperatively, X-ray and CT showed that the fusion instrument and bone have been fused; MRI showed the disappearance of syringomyelia (Fig. 8A-D). The JOA score of the cervical spine was 17 points in 21 months following the surgery (Fig. 9A-D).



Figure 5. A–D, The blue part of the surface in contact with the bone is to be printed in 3D microwells (A–C); the match between the 3D-OCF and the 3D model (E). 3D-OCF = 3D printing occipitocervical fusion instrument.



Figure 6. A-D, Intraoperative matching and fixing: trial matching (A); installation of 3D-OCF, guide sleeve (B); drilling (C); fixing (D).



Figure 7. A–D, X-rays of the cervical spine 2 days after operation (A, B); CT at 10 days after operation shows reduction of the atlantoaxial vertebrae and enlargement of the occipital foramen (C); MRI at 10 days after operation shows reduction of spinal cavity (D).



Figure 8. A–D, X-ray and CT showed fusion of 3D-OCF and bone interface (A, B, at 6mo); MRI showed disappearance of spinal cavity (C, D, at 6mo).



Figure 9. A–D, X-ray and CT showed fusion of 3D-OCF and bone interface at 21 mo after operation.

### 4. Discussion

Upper cervical surgery is one of the most challenging problems in the field of spine. Most patients with upper cervical spine deformity or trauma have upper cervical instability, which is called high-risk upper cervical vertebra in clinic. The internal fixation of high-risk upper cervical surgery mainly includes anterior transpharyngeal reduction and plate internal fixation, posterior reduction screw-rod internal fixation, and anterior oropharyngeal release combined with posterior screw-rod internal fixation. There are numerous complications in anterior oropharyngeal surgery. Wu et al<sup>[7]</sup> reported that anterior oropharyngeal surgery may be complicated by mixed incision infection, affecting vocalization, tongue edema, retropharyngeal hematoma, and cerebrospinal fluid leakage.

However, the complexity of the anatomical structure adjacent to the pedicle of the upper cervical spine, and the individual differences make it difficult and risky to place pedicle screws through the posterior approach. When establishing the channel during the operation, even the experienced spine surgeons are difficult to quickly and accurately complete the placement of the upper cervical pedicle screw. If the screw placement direction cannot be well controlled or the screw placement position is inaccuracy, it will not only cause spinal cord, vertebral artery, and dura mater injury, but also lead to internal fixation strength decline or even failure, increased incidence of postoperative complications.<sup>[8,9]</sup> Ebraheim et al<sup>[10]</sup> indicated that the accuracy rate of cervical pedicle screw placement was only 71.28%. What's more, there is report of epidural hematomas in the posterior cranial fossa.<sup>[11]</sup> The C-arm X-ray machine is used to repeatedly guide the screw placement in traditional surgery; there are some other problems such as high surgical risk, prolonged operation time, and low efficiency. Various individuals have anatomical differences in the upper cervical spine, thus unified screw placement cannot be selected. The large surgical navigation equipment is expensive, which is not conducive to popularization. Therefore, the internal fixation of posterior screw-rod system of upper cervical spine cannot be carried out routinely in primary hospitals.

The internal fixation of the upper cervical spine with posterior screw-rod system is difficult, and the bone fusion is a critical step. If the upper cervical vertebra is fixed without osseointegration and the screw-rod system is tired over time, which will lead to the possibility of internal plant failure or even break.<sup>[12,13]</sup> Kukreja et al found 2 patients with pseudoarthrosis due to poor osseous fusion in 36 patients with upper cervical surgery.<sup>[14]</sup>

Posterior bone grafting usually comes from the patient's autogenous ilium or the spinous process and lamina of surgical vertebral body, which undoubtedly brings iatrogenic trauma to the patient or further reduces the stability of the cervical vertebra. The most common complications in the bone removal are hematoma, pain, and infection.<sup>[15]</sup> Therefore, it is worthy of research to find a new occipitocervical fusion instrument. Recently, with the development of rapid prototyping technology, 3D printing has been successfully used in neurosurgery, cardiothoracic surgery, maxillofacial stomatology, and other fields, especially in orthopedics.<sup>[16,17]</sup> 3D printing assisted intraoperative operations and customized implants have become hot spots in the development of spinal surgery.<sup>[18]</sup> We combine 3D printing technology with modern imaging and the principle of computer 3D reconstruction reverse engineering principles, and used titanium powder as a material to print individualized posterior cervical spinal fusion instrument with screw guide holes. 3D-OCF integrates the functions of fixation, nailing, and fusion, has revolutionized the previous concept, changing the traditional occipitocervical fusion screw-rod split system to an integrated system according to the anatomical structure of patients. The fusion instrument combined with personalized guidance technology can observably reduce the number of intraoperative fluoroscopy and operative time, significantly improve the efficiency and safety of screw placement in upper cervical surgery.<sup>[19]</sup> Additionally, this technique can also accurately implant screws into other structures, such as spinous process, pyramidal plate. Thayaparan et al $^{[20]}$  also developed a 3D printing customized integrated occipital-cervical fusion system and achieved satisfactory clinical treatment outcomes in upper cervical surgery. However, there is no porous structure on the inner surface of this fusion system, and autogenous iliac bone is needed for bone graft fusion. The bone-contacting surface of our designed fusion device adopts the porous structure of bionic trabeculae, which can contribute to bone growth. Moreover, we verified that the porous titanium implant is beneficial to bone growth through animal experiments, and

screened out the range of porosity gradient conducive to bone growth on the surface of cortical bone.<sup>[21]</sup> This porous structure enables the fusion instrument to obtain strong fixation, which can avoid the implantation of autologous bone during operation, reduce iatrogenic trauma, achieve rapid bone fusion, and reduce the risk of broken screws and rods.<sup>[22]</sup>

# 5. Conclusion

Individualized 3D-OCF fully based on individual anatomy, which can improve the safety and accuracy of surgery, reduce the operative time and the number of fluoroscopy. Our study provides a novel surgical approach for the challenging upper cervical surgery.

# Acknowledgments

The authors acknowledge AK Medical Company for its academic support.

# Author contributions

GQ N conceived of the design of the study. GQ N, H C, G Z, H N, LT L, JQ Z, and JH D participated in the operation. GQ N, C L and QK Z performed the 3D model printing. GQ N, JZ B, and LT L finished the manuscript. GQ N and JZ B participated in the revision of the article. All authors read and approved the final manuscript.

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