ANTERIOR STABILIZATION OF C1–C4 VERTEBRAE AFTER TRANSORAL REMOVAL OF A GIANT CELL TUMOR OF C2–C3 VERTEBRAE

A.N. Shkarubo[1], A.A. Kuleshov[2], I.V. Chernov[1], N.A. Eskin[2], G.N. Berchenko[2], M.S. Vetrile[2], I.N. Lisyansky[2], S.N. Makarov[2]

[1]N.N. Burdenko National Scientific and Practical Center for Neurosurgery, Moscow, Russia

[2]N.N. Priorov National Scientific Center of Traumatology and Orthopaedics, Moscow, Russia

A clinical case of a single-step transoral removal of C2 and C3 vertebral bodies with subsequent stabilization of the C1—C4 spinal segment through the same approach (with additional submandibular approach to insert screws into the C4 vertebra body) without dissection of the mandible and tongue is presented. The first experience of anterior stabilization of the C1—C4 spinal segment using a custom-made instrumentation system demonstrated its effectiveness. It was shown that, taking into account the length of the cervical spine resection, the anterior stabilization with a custom-made plate should be combined with standard methods of posterior fixation. Innovative surgical technologies allow optimizing the surgical technique of cervical spine stabilization and provide earlier rehabilitation of a patient.

Key Words: transoral approach, anterior stabilization, giant cell tumor.

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Giant cell tumors are benign, but locally aggressive tumors [1]. They most frequently occur in young adults between 20 and 40 years of age [2–4]. There are data on a significant incidence of giant cell tumors for the Chinese population, up to 20 % of all bone tumors [4, 5]. Although giant cell tumors are the most widespread primary bone tumors, spine tumors are of rare occurrence [6]. Tumor morbidity in the mobile part of the spine (above the sacrum) varies between 1.4 and 9.4 %, meanwhile the cervical spine involvement is even less [7]. Despite benign nature of tumor, it can be locally aggressive and have a metastatic potential [8].

Curettage is the most widespread surgical approach to treatment of giant cell tumors of the spine, however incomplete removal of a tumor usually leads to recurrent tumor and metastasis [9].

It is known that en bloc resection in the cervical spine is impossible because of the necessity to preserve the vertebral arteries and nerve roots innervating the upper extremities [10]. A wide resection is theoretically aimed to prevent local recurrence, but at the same time it increases risks of such complications as hematoma, functional deficit, fistulas, infections, and instability of the atlantoaxial joint, when the C1 or C2 vertebrae are injured [11].

Due to the anatomic peculiarities of the upper cervical spine, anterior approach is the mostly widespread and justified for the C2 and C3 vertebrae resection [12, 13], but in the majority of cases, the instability of the atlantoaxial joint arises after similar surgeries.

Nowadays, a transoral decompression followed by posterior stabilization of the upper cervical spine is the accepted treatment approach of various diseases associated with the atlantoaxial instability and anterior compression of the brain stem [14–20]. However, the necessity to turn the patient and the additional approach for posterior stabilization extend the surgery time and increase its traumaticity [21]. Now it is possible to carry out anterior stabilization of the atlantoaxial joint as a single-step surgery without any intraoperative turn of the patient because of the development of surgical technologies [22–24].

Stulik et al. [25] described a case of a total spondylectomy of the C2 vertebra due to solitary metastasis of thyroid adenocarcinoma. In the first step, the authors applied the posterior fixation of C1–C4. Subsequently, they removed the C2 vertebral body with the tumor through the anterior transmandibular approach (the mandible was dissected) and carried out spinal fusion of C1–C3 with a shaped SynMesh cage. It is necessary to underline that this type of surgery is highly traumatic because of the dissection of mandible and tongue.

The present paper describes a case of a transoral removal of the tumor with a single-step transoral stabilization of the C1–C4 spinal segment (through the additional submandibular approach) using a custom-made expanding stabilization system without mandible dissection.

Patient P., 57 years old, with a giant cell tumor of the C2 and C3 vertebral body and complaining of local pain syndrome in the area of the neck and restricted mobility in the cervical spine was admitted to hospital for a surgical intervention.

According to the patient's medical history, the pain in the cervical spine appeared for the first time in January, 2016. MRT and HCT of the cervical spine revealed destruction of the C2 and C3 vertebral bodies (Fig. 1).

Taking into account an extremely high risk of the development of pathologic fracture of the C2 and C3 vertebrae and appearance of a rough neurological deficit with a threat to life of the patient, the first step of the surgical treatment was carried out on 07.08.2017, including laminectomy of C3, spinal cord decompression, occipitospondylodesis with the use of metal constructs and autobone graft.

The postoperative period was without complications. The control CT showed a satisfactory state of the stabilizing system (Fig. 2).

Three months later, the patient was admitted to the hospital for the second step of the surgical treatment. A customized extending stabilizing system was prepared using mathematical modeling as a preparation stage for the surgery (Fig. 3). The tumor of the C2–C3 vertebrae was resected through the transoral and submandibular approaches, as well as anterior decompression of the spinal cord along the length of C1–C3, anterior atlantosubaxial stabilization of the C1–C4 vertebrae using the customized stabilizing system were performed on 08.11.2017 (Fig. 4).

Fig. 1
HCT, MRI, and HCT 3D reconstructions before the surgery visualize the C2 vertebral body injury, nearly total destruction of the C3 vertebral body, and spinal stenosis

The surgical technique. The patient was in the supine position. The tracheostoma was installed. A 5 cm skin incision was made on the anterior surface of the neck on the right, by 3 cm lower the inferior border of the mandible. The cervical fasciae were dissected. The cervical vessels were shifted laterally, and the cervical organs were shifted medially. The C4, C3 vertebral bodies and the inferior border of the C2 vertebra were skeletonized; a pronounced height loss of the C3 vertebral body was noted, as well as the presence of pathological tissue. The C3–C4 disc was resected, while the C3 vertebral body was resected partially. Hemostasis. The wound was swabbed. The mouth opener was placed. The approach to the anterior surface of the C1–C3 vertebral bodies was performed. The C2 vertebral body destruction and nearly complete destruction of the C3 vertebral body were revealed. The pathologically changed C2–C3 vertebral bodies, as well as the changed tissue of the odontoid bone were resected using a high-speed bone drill, bone scrapers, laminotoms, and discotoms. The tumor was heavily vascularized. It was removed completely. Then the dura mater was exposed for 4 cm, its pulsation was well-marked. The assembled customized extending metal plate was inserted into the wound through the transoral approach, the plate was extended from the C1 to C4 vertebrae. The temporary fixation of the lower pole to the C4 vertebra was performed through the submandibular approach. The radiological control showed a correct position of the system (Fig. 5). Finally, the plate was fixed with metal screws in the lateral masses of C1 and in the C4 vertebral body (by two screws in each, respectively). The upper screws were inserted into the lateral masses of the C1 vertebra transorally, and the lower screws were inserted into the C4 vertebral body through the submandibular approach. The soft tissues were closed layer by layer.

The histopathological examination showed a giant cell tumor with single pathologic mitoses and single patches of atypical osteoid (Fig. 6).

The early postoperative period was without complications. The control X-ray patterns and CT showed the correct position of the system (Fig. 7). The

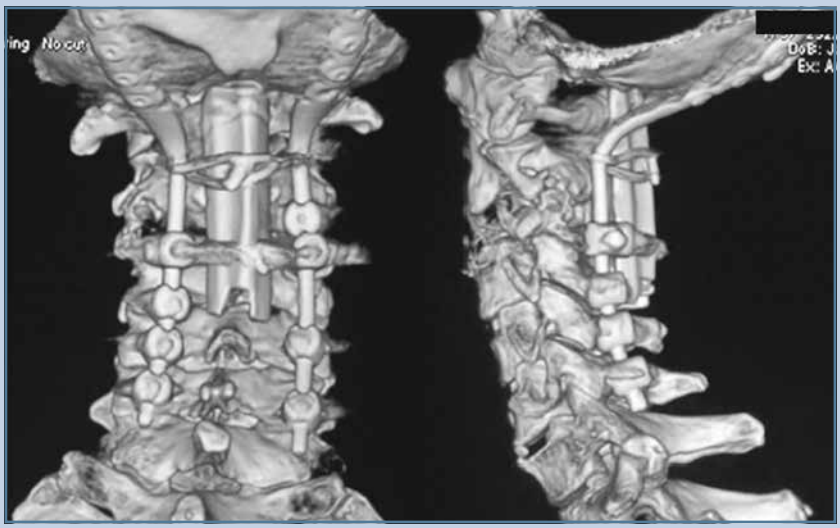

Fig. 2
HCT 3D reconstruction of the patient P., 57 years old, after the first surgery (the system for occipitospondylodesis is installed)

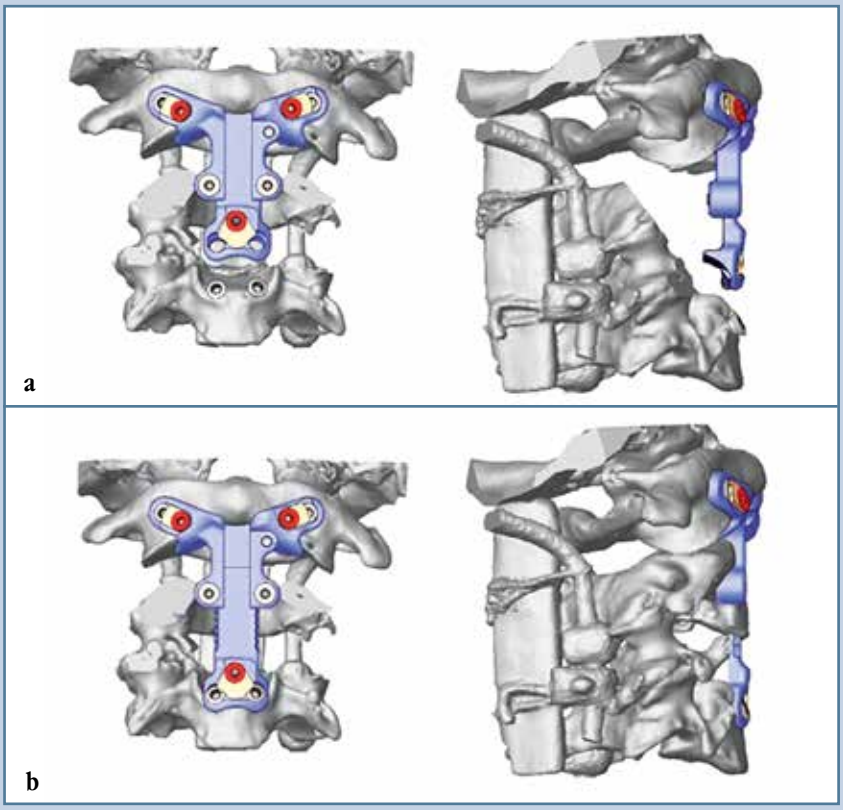

Fig. 3
Modeling of bone structures of the C1–C4 spinal segment of the patient P., 57 years old, the system for anterior stabilization of C1–C4 is installed: **a** – anterior and lateral views of the assembled system; **b** –anterior and lateral views of the system in the extended position

patient was discharged in satisfactory condition.

Discussion

Giant cell tumors are widespread benign primary bone tumors, which are well-known by their locally aggressive behavior and tendency to recurrence [26]. Despite the benign nature of their majority, 5 to 10 % of tumors are malignant and aggressive [27, 28]. The aggressivity of tumors leads to local recurrence in 7–75 % of patients [29].

For the time being, there is no standard surgical technique for treating giant cell tumor of the cervical spine, especially in the C1 and C2 vertebrae because of the anatomic peculiarities and technical difficulty of interventions to the upper cervical spine. Traditionally, the surgical treatment of such tumors includes a wide en-bloc resection, intracellular curettage [30, 31], and X-ray therapy [32]. Occipitospondylodesis is the most widely used stabilizing surgery for the C2 vertebra resection. Without doubt, the function of the atlanto-occipital joint is lost after such fixation [33]. It is a vital problem to elaborate alternative methods for stabilizing the upper cervical spine in diseases located in the region of the craniovertebral junction [34].

The treatment of patients with atlantoaxial instability caused by various diseases in the region of the skull base and craniovertebral junction, which are combined with ventral compression of the brain stem, is still too complicated and controversial [35].

For the first time, Schmelzle et al. [36] described anterior stabilization of the atlantoaxial joint in 1987. Anterior stabilization proved its safety in the course of biomechanical and clinical studies [30, 37–39]. However, the use of the Harms plate was limited at that period of time because of the problem: the screws untwisted themselves too often [37, 39].

At present, application of transoral atlantoaxial reduction plates is one of

Fig. 4
The system for the C1–C4 vertebrae anterior stabilization: **a** – the assembled system; **b** – the system in the extended position; **c** – the system in the extended position is on the stereolithographic model of the upper cervical spine of the patient P., 57 years old

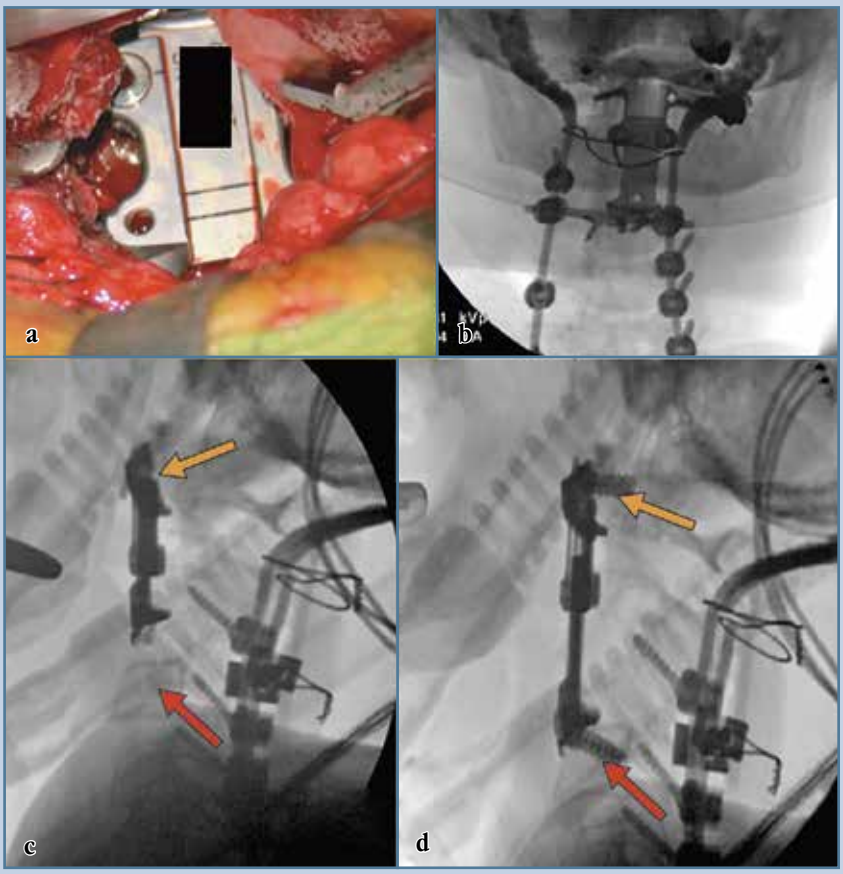

Fig. 5
Inserting the system for anterior stabilization of the C1–C4 vertebrae of the patient P., 57 years old: **a** – the intraoperative image shows the insertion through the transoral approach on the anterior surface of the C1–C4 vertebrae; **b, c** – the X-ray patterns show the assembled system in frontal and lateral projections: the yellow arrow points to the C1 vertebra, the red arrow points to the C4 vertebra; **d** – the X-ray pattern shows the system in the extended position in lateral projection: the yellow arrow points to the C1 vertebra, the red arrow points to the C4 vertebra, the screws are untwisted

the most commonly used systems of stabilization. Transoral atlantoaxial anchored cage is also known [30].

A lot of studies proved the efficiency of anterior stabilization using various systems [22, 23, 24, 35, 40, 41]. A system for anterior stabilization of the C1–C4 vertebrae was elaborated [42]; its joining surface is absolutely congruent to the anterior surface of the C1–C4 spinal segment of a particular patient; this system has a supporting element with additional support points in the inferior articular surface of the C1 and C4 upper vertebral body, the system also includes locking elements, which do not let screws untwist. This is a unique system: it is inserted transorally as an assembled unit, and then it is extended to the desired size intraoperatively and fixed. The stabilizing system is produced using mathematical modeling of the craniovertebral segment of the spine of a particular patient. We studied biomechanical efficiency of anterior and posterior stabilizing constructs and demonstrated high efficiency of anterior stabilization of the atlantoaxial joint [43].

The presented clinical example demonstrates a possibility of a single-step removal of a tumor of the C2, C3 vertebral bodies through the transoral and submandibular approaches with the subsequent stabilization of the C1–C4 spinal segment through the same approach without dissection of mandible and tongue. In this case, taking into account the length of the resection of anterior supporting structures of the cervical spine, the anterior fixation should be combined with posterior stabilization.

The study had no sponsorship. The authors declare no conflict of interest.

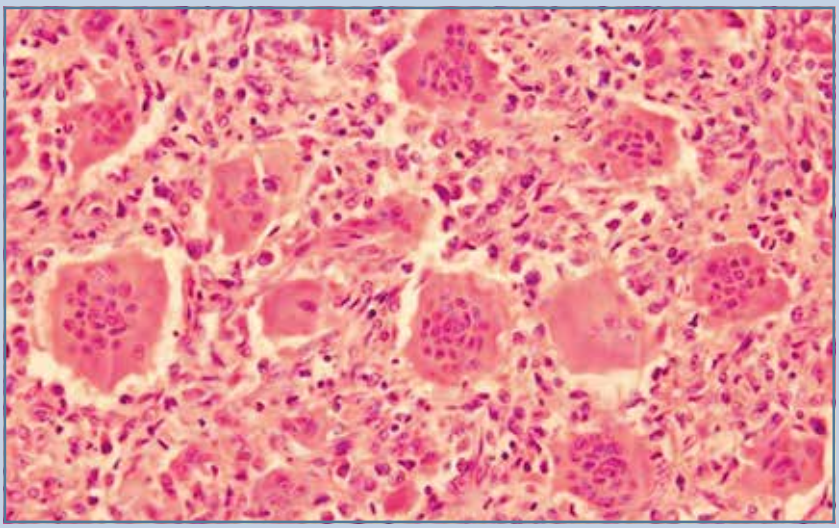

Fig. 6
Numerous giant polynuclear osteoclast-like cells with mononuclear cells located among them; staining with H&E, magnification of 400x

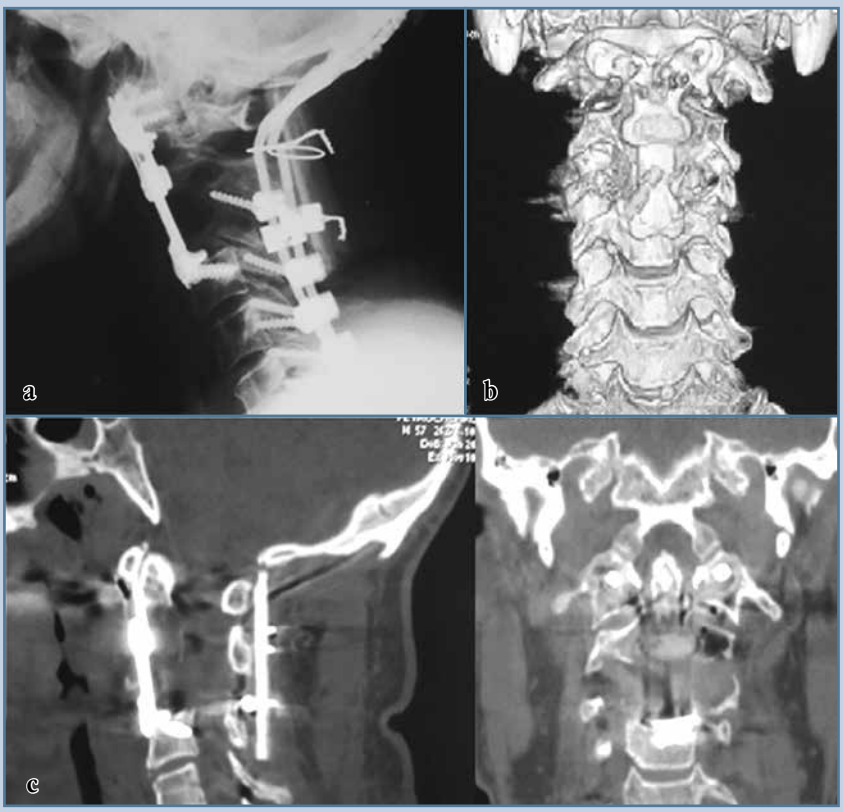

Fig. 7
Survey radiograph of the patient P., 57 years old, after the surgery: in lateral projection **(a)**, CT 3D reconstruction **(b)**, and in sagittal and frontal projections **(c)** shows: the correct position of the system and the total removal of the pathologic tissue

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Address correspondence to:
Shkarubo Aleksey Nikolaevich
4-ya Tverskaya-Yamskaya, 16, Moscow, 125047, Russia,
N.N. Burdenko National Scientific and Practical Center for Neurosurgery,
ashkarubo@nsi.ru

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Aleksey Nikolayevich Shkarubo, DMSc, leading researcher, Neurooncological Department No. 8, N.N. Burdenko National Scientific and Practical Center for Neurosurgery, 4-ya Tverskaya-Yamskaya str., 16, Moscow, 125047, Russia, ORCID: 0000-0003-3445-3115, ashkarubo@nsi.ru;
Aleksandr Alekseyevich Kuleshov, DMSc, leading researcher, head of vertebrology department, N.N. Priorov National Medical Research Center of Traumatology and Orthopaedics, Priorova str., 10, Moscow, 127299, Russia, cito-spine@mail.ru;
Ilya Valeryevich Chernov, postgraduate student, neurooncological department No. 8, N.N. Burdenko National Scientific and Practical Center for Neurosurgery, 4-ya Tverskaya-Yamskaya str., 16, Moscow, 125047, Russia, ORCID: 0000-0002-9789-3452;
Nikolay Aleksandrovich Eskin, DMSc, Prof., deputy director for science, N.N. Priorov National Medical Research Center of Traumatology and Orthopaedics, Priorova str., 10, Moscow, 127299, Russia, dissovet@cito-priorov.ru;
Gennady Nikolayevich Berchenko, DMSc, Prof., head of pathological anatomy department, N.N. Priorov National Medical Research Center of Traumatology and Orthopaedics, Priorova str., 10, Moscow, 127299, Russia, berchenko@cito-bone.ru;
Marchel Stepanovich Vetrile, MD, PhD, senior researcher, vertebrology department, N.N. Priorov National Medical Research Center of Traumatology and Orthopaedics, Priorov str., 10, Moscow, 127299, Russia, cito-spine@mail.ru;
Igor Nikolayevich Lisyansky, MD, PhD, physician of vertebrology department, N.N. Priorov National Medical Research Center of Traumatology and Orthopaedics, Priorova str., 10, Moscow, 127299, Russia, cito-spine@mail.ru;
Sergey Nikolayevich Makarov, MD, PhD, physician of vertebrology department, N.N. Priorov National Medical Research Center of Traumatology and Orthopaedics, Priorova str., 10, Moscow, 127299, Russia, ORCID: 0000-0001-8364-5223, cito-spine@mail.ru.