LONG-TERM RESULTS OF TREATMENT OF A PATIENT WITH BASILAR INVAGINATION COMPLICATED BY DISTAL KYPHOSIS AND COMPRESSIVE ISCHEMIC CERVICAL MYELOPATHY: A CLINICAL CASE AND A BRIEF LITERATURE REVIEW

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Objective. To present a clinical case of surgical correction of a craniovertebral anomaly complicated after 8 years by distal junctional kyphosis, stenosis, antelisthesis of the C5 vertebra and compressive ischemic cervical myelopathy in the C5—C6 segment.
Material and Methods. When treating a 56-year-old patient with multiple anomalies of the craniovertebral region, differentiated surgical technologies were consistently used due to the development of late complications. The sequence and rationale for surgical decision making is described.
Results. Initially, the patient underwent transoral decompression and posterior occipitocervical fixation, and after 8 years - reinstallation of the system with distal extension of the instrumentation zone to the C7 vertebra with indirect posterior decompression of the spinal cord, anterior discectomy with direct decompression and cage fixation at the C5—C6 level. Regression of myelopathic syndrome, correction of orthopedic status and significant improvement in functional status were achieved.
Conclusion. A rare clinical observation demonstrates a combination of basilar invagination with assimilation of the atlas, which has provided rationale for two-stage surgical treatment in one surgical session (1st stage — transoral resection of the dens and 2/3 of the C2 vertebral body with anterior decompression of the spinal cord, and 2nd stage — occipitocervical fixation). The use of extended systems in this case caused the development of a clinically significant syndrome of the distal adjacent level, which required repeated surgical treatment after 8 years.
Key Words: craniovertebral junction, occipitocervical fixation, occipitospondylodesis, adjacent level syndrome, distal junctional kyphosis, cervical myelopathy.
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The craniovertebral junction (CVJ) is a sophisticated anatomical and biomechanical system which boundaries presented by bone, ligamentous, vascular, and neural structures that extend from the lower third of the clivus and condyles of the occipital bone to the C2–C3 intervertebral disc. The CVJ provides head movement with maximum amplitudes in all three planes, although it is biomechanically susceptible, which is true for all junctional areas [1–3]. The main pathological conditions (injuries, tumors of supporting structures and intradural neoplasms, abnormalities, infectious and inflammatory processes, and genetic diseases) localized in this region can be represented by one or more dominant clinical syndromes or a combination of them: compression, instability, deformities, and vascular and liquor dynamic disorders [4–7]. Most pathological processes localized in the CVJ result in instability requiring determination of its elimination.

Nowadays, there are two main approaches to correction of instability and compression of the CVJ: occipitocervical fixation (OCF) and short-segment Goel – Harms techniques [2, 3, 8, 9].

OCF is a widely used surgical technique to correct the instability of the CVJ of various origins [1, 9–12]. Before the advent of modern instrumentation and technologies, fixation was performed in various ways, including using wire and autogenous bone grafts, which required long-term postoperative wearing of a rigid cervical collar or halo ring at a sufficiently high frequency of non-union [10–12]. Modern OCF implies rigid intraoperative stabilization with an occipital plate, polyaxial screws, and rods, which enable repositioning and do not require additional postoperative immobilization of the cervical spine [9, 10, 12, 13].

OCF may be complicated by a number of serious conditions: injuries to neural structures, large arterial vessels, venous plexuses, failure and fracturing of the structure, and pseudoarthrosis [10, 12, 13]. Their incidence varies from 10 to 52 % [10, 12, 13]. The long-term outcomes of OCF have been less studied in terms of the development of adjacent segment disease and the formation of distal junctional kyphosis [10, 13].

The objective is to present a clinical case of surgical correction of a craniovertebral abnormality complicated after 8 years by distal junctional kyphosis, stenosis, antelisthesis of the C5 vertebra and compressive ischemic cervical myelopathy in the C5–C6 segment.

Study design: clinical observation with long-term follow-up and a brief literature review.

First visit

Male patient S., 56 years old, at the time of the first visit in 2015, had a strong-built, and stood 182 cm tall, weighed 110 kg. He lived an active lifestyle, served his compulsory military service in the airborne troops. After demobilization, he worked as a bricklayer at a construction site, where at the age of 53 he was injured that provoked the onset of the disease: during building and construction work, a beam weighing about 10–15 kg fell on his helmet. A few months later, burning pains appeared in the occipitocervical region, which prompted the patient to seek medical advice. The patient underwent outpatient care with a diagnosis of a degenerative spine disease. The imaging studies had not been performed.

The first manifestations of ataxia appeared after one year, against the background of the preservation of mentioned symptoms, but the patient continued his work activities. An impaired sensation occurred mainly in the upper extremities associated with the gradual progression of ataxia. Three years after the primary symptoms appeared (2015), the physical function decompensated: quadriparesis and bulbar palsy (choking when eating solid food) developed associated with a significant progression of ataxia. The patient stopped not only his work activities but also serving himself in everyday life. During this period, he was referred to a neurosurgeon for the first time.

At the time of admission to the hospital, the physical functional status was as follows: quadriparesis according to the European Myelopathy Scale (EMS) – grade II (10 points); neck pain – 9/10 points according to the VAS; pronounced bulbar palsy – dysphagia and choking.

The following multiple CVJ abnormalities were revealed in the patient: basilar invagination, Chiari type I malformation, atlas assimilation with the development of cervicomedullary compression (Fig. 1, 2). The main indices and criteria describing the angulometric indicators of the CVJ that resulted in cervicomedullary compression, are given in Table 1.

Clinical presentation before the first surgery

The severity of the patient's condition at the time of admission depended on cervicomedullary compression. According to spiral computed tomography and MRI findings, the patient suffered from clinical manifestations of craniovertebral instability associated with multiple congenital abnormalities. Meanwhile, the nature of the abnormalities (mainly blocking) indicates not so much the initial instability but rather a stable, long-lasting compensated basilar impression with functional decompensation, apparently triggered by injury. Clinical symptoms indicate that degenerative changes in the C3–C4 segment are not the cause for the manifestation of pathology. Consequently, the primary aim of surgical treatment at this stage was decompression of the spinal cord and the lower parts of the trunk in the craniovertebral junction.

The combination of basilar invagination with the atlas assimilation determined the choice of two-stage surgical treatment in one session: stage 1 – decompression (transoral resection of the dens and 2/3 of the C2 vertebral body); stage 2 – OCF (Fig. 3). Considering the gross clinical manifestations of compression of the brain stem, it was decided less hazardous for the patient to perform decompression at the first stage, since there was a high risk of aggravation of neurological disorders during the fulfilment of the second stage (reduction of dislocation followed by OCF). Technical peculiarities of the OCF: the C2 vertebra was not included in the fixation zone due to transoral resection of the dens and 2/3 of the C2 vertebra body; in total 3 pairs of screws were placed through the lateral masses of the C3, C4 and C5 vertebrae.

A rapid recovery of the patient's neurological functions was observed in the postoperative period. On the 10th day, the patient moved independently, could serve himself in everyday life, and his bulbar palsy regressed. Recovery of working ability was observed 6–7 weeks after surgery (Table 2); the patient returned to his previous duties.

For seven years after the surgery, the patient led a full life and worked as a high rigger. In September 2022, he experienced pain in the cervical and occipital regions. Hypoesthesia started to develop in the extremities in March 2023 and manifested in a "stocking and glove" distribution, extremity weakness and unsteadiness of gait. The patient stopped his work activities and then lost the opportunity to serve himself in everyday life. At the time of the second admission, at the age of 64 (May 2023), he had the same physical parameters: height 182 cm, weight 115 kg.

Physical function at the time of admission to the hospital: European Myelopathy Scale (EMS) – grade II (11 points); neck pain – 7/10 points according to the VAS.

The MRI findings revealed degeneration of level adjacent to structure with formation of a myelophthisic area in the spinal cord at the C5–C6 level (Fig. 4). This suggests that the spinal cord had experienced transient compression at the C5–C6 vertebral level. According to the spiral computed tomography findings, the following features were detected: the left rod hooked with the distal end to the C6 vertebral arch and did not allow the entire OCF structure to shift, resulting in a pressure ulcer (Fig. 5).

Clinical presentation before the second surgery

At the time of repeated treatment, severity of patient's condition was presented as the pathology of the distal (in relation to the primary instrumental fixation) contact segment, which was characterized by segmental instability, the formation of distal junctional kyphosis (possibly due to the mechanical pressure of the distal ends of the OCF structure) and the development of cervical myelopathy. Thus, the aim of the surgery was spinal cord decompression and stabilization of the C5–C6 segment. Nevertheless, it seemed impossible for us to implement decompression and stabilization through the anterior approach without preliminary eliminating the pressure of the distal ends of the rods of the OCF structure from behind on the C6 vertebral arch. We decided to perform the surgery in 2 stages.

The first stage consisted of partial reinstallation of the elements of the OCF system (installation of additional polyaxial screws in the C6 and C7 vertebrae and extension of the parallel rods of the structure to the C7 vertebra with indirect spinal cord decompression). During the surgery, a fracture of the right rod was found under the nut of the C3 polyaxial screw that was not invisible in the spiral computed tomography scan. It was impossible to place a screw into the facet of the C6 vertebra on the left due to its fracture along the line of the pressure ulcer associated with the pressure of the distal end of the rod. The second stage (5 days after): anterior discectomy with spinal cord decompression and cage fixation to create a bone block between the C5 and C6 vertebrae. As a result, it was possible to achieve indirect decompression and stability of OCF (Fig. 6, 7).

In the early postoperative period, the patient showed regression of manifestations of myelopathic syndrome (grade III, 13 points according to the EMS). The neck pain had completely stopped; the manifestations of ataxia decreased. On the 10th day after surgery, the patient moved independently, could serve himself in everyday life. According to the control spiral computed tomography scan, the sagittal balance of the patient's trunk was restored (Table 3, Fig. 7).

Discussion

The techniques of short segment stabilization in the surgical treatment of CVJ pathology have been growing in popularity in the last two decades [2, 3, 8, 15]. Their advantage, in comparison with extended occipitocervical fixation systems, is the preservation of mobility of the superjacent (C0–C1) and subjacent

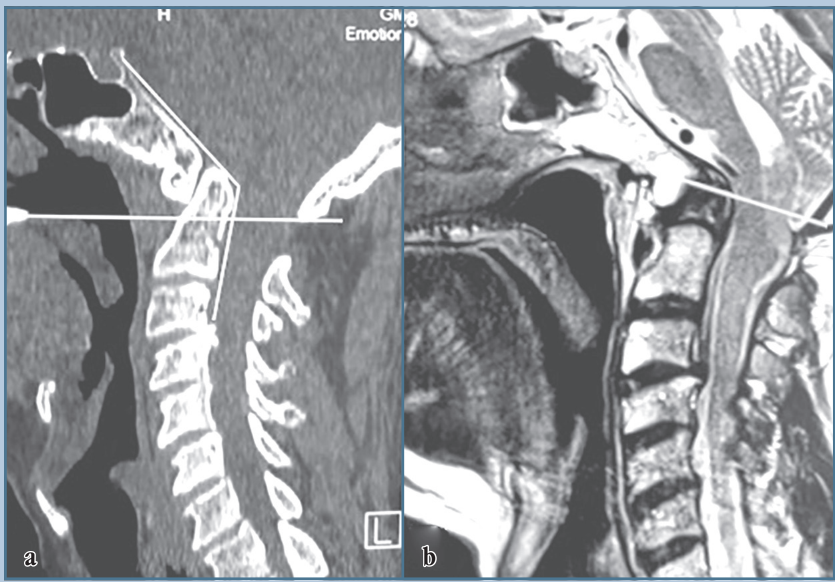

Fig. 1
SCT **(a)** and MRI **(b)** of patient S. in the sagittal plane before surgery: **a** – Wackenheim clivus-canal angle and Chamberlain line; **b** – McRae line; location of the dens of the C2 vertebra in the great occipital foramen with compression of the brain stem

Fig. 2
SCT of patient S. in sagittal **(a)**, coronal **(b)** and axial **(c)** planes before surgery: assimilation of the atlas **(a, b)**, bone cyst in the clivus **(c)**

Table 1
Measurements of ratios (angles and lines) in the craniovertebral junction at the first admission of patient S.

Criterion	Value
Wackenheim clivus-canal angle	135°
McRae line	Superjacent dens
Chamberlain line	Superjacent dens
Wackenheim line	Superjacent dens

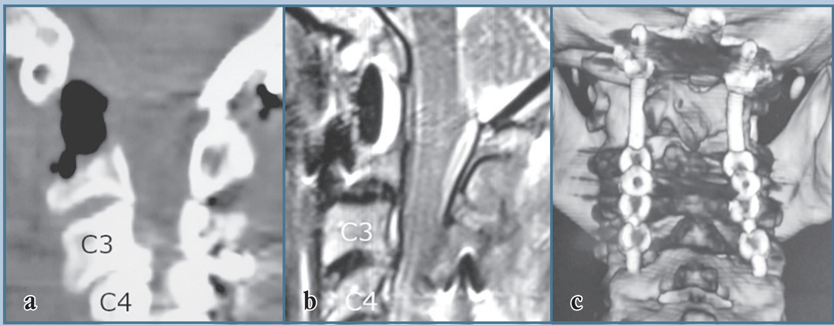

Fig. 3
SCT **(a)** and MRI **(b)** of patient S. in the sagittal plane after the first surgery, 3D reconstruction **(c)**: **a** – resection of the dens and 2/3 of the C2 vertebral body; **b** – cervicomedullary decompression; **c** – occipitocervical fixation (C0–C3–C4–C5)

Table 2
Dynamics of the functional status of patient S.

Scale	Initially	10 days after	6 weeks after
Nurick, grade	V	I	0
EMS, points	10 (EMS II)	15 (EMS I)	18 (normal)
VAS, points	9	3	0

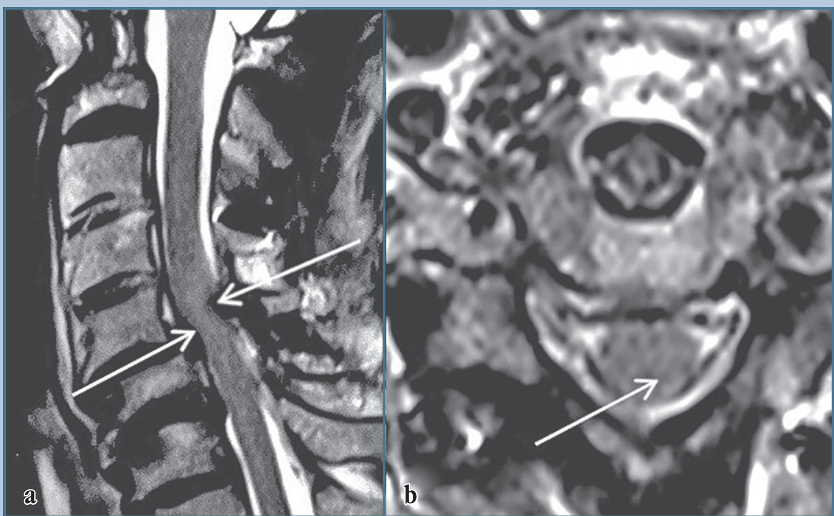

Fig. 4
MRI of patient S. in sagittal **(a)** and axial **(b)** planes before surgery: at the second admission, C5 antelisthesis and spinal canal stenosis in the C5–C6 segment with compression of the spinal cord and the formation of a focus of altered MR signal at the level of C5–C6 with a decrease in anterior height of C6 body, DJS with instability and secondary myelopathy

segments, while these techniques do not have statistically significant differences in functional outcomes [16, 17]. In turn, atlantoaxial fixation (C1–C2) gives a considerable advantage in the range of motions due to the preservation of mobility in the C0–C1 segment [16–18]. There is little data in the literature on the degeneration of the adjacent level and even less on the development of distal junctional kyphosis after the use of OCF [16, 17].

Nowadays, both approaches are used in CVJ surgery. The OCF system is a forced but justified procedure used when there is no stable point for the implantation of a polyaxial screw of short-segment fixation (bone destruction, osteoporosis, abnormal vertebral artery permeability, congenital anatomical abnormalities of spine regions, etc.) [16–19]. Our case illustrates a similar situation: the assimilation of the atlas with the occipital bone makes no sense to preserve the unfixed C0–C1 segment, and the underdeveloped lateral mass of C1 is not a reliable support for the installation of a polyaxial screw (Fig. 1, 2).

Some articles present data on the long-term outcomes of OCF from the perspective of an adjacent segment disease with the development of distal junctional kyphosis [20, 21]. OCF accelerates the phenomenon of degeneration below the fixation area and results in its formation. Revision surgery (surgery of consequences) after OCF has its own technical peculiarities and is much more complicated in deciding on the techniques and sequence of surgical stages of treatment [21–23].

Adjacent segment disease is a well-known phenomenon in spine surgery. In the case of anterior cervical fusion, its frequency is about 3 % per year with no significant differences in the surgical techniques [24–26]. In the case of posterior cervical fusion, radiological signs of the disease account for 3.4 % in the first year and 5.9 % in the second year after surgery, and with a long-term analysis of outcomes, it will occur in 20–30 %. Clinical manifestations in this case amount to 30–60 % [24–26]. In the case of OCF, there is not much data on the adjacent segment disease in the literature. According to Zileli and Akintürk [13], it occurs in 7 % of cases.

The development of distal junctional kyphosis is quite a big problem in the treatment of deformities of the spine in all its departments, including the CVJ. According to Passias et al. [23], the incidence of distal junctional kyphosis in the cervical spine in various pathologies and the extent of surgeries can reach 20–25 % [21, 23].

The literature provides data on the impact of risk factors for the development of distal junctional kyphosis in patients after surgical correction of cervical spine deformities: demographic, clinical and neurological, radiological, and surgical [21, 23]. The literature's data on risk factors for the development of distal junctional kyphosis in the cervical spine were examined without considering the cervical spine fixation to the occipital bone with a plate. We have not found any separate papers on OCF.

According to modern studies, the more segments of the cervical spine are fixed and the larger the extent of bone resection, the greater possibility of development of adjacent segment disease and distal junctional kyphosis. The use of transition rods (from the cervical to the thoracic spine) also enhances the risk of distal junctional kyphosis [21, 23, 26]. Occipitospondylodesis technique presupposes a fixed blockage of the skull (squama occipitalis) and the cervical spine (facet joints or pedicles), which is an aggravating factor for the development of distal junctional kyphosis [21, 23, 26].

In the presented clinical case, during the first patient's admission, decompression was first required using one of two options: either direct decompression (transoral/transnasal resection of the dens of C2) or indirect decompression using the Goel technique [2, 3, 27]. We performed an anterior decompression of the spinal cord though the transoral approach, and the use of a long-segment OCF system was a forced measure due to the presence of an abnormality and degeneration of the C3–C4 segment. The implemented surgical treatment strategy

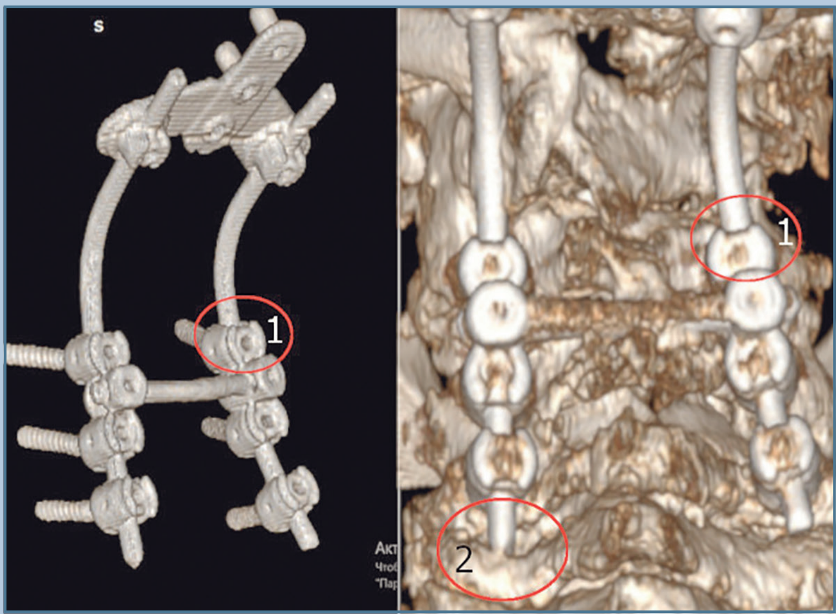

Fig. 5
SCT (3D reconstruction) of patient S. before surgery during the second admission: a fracture of the structural rod under the nut *(1)*, which was discovered only intraoperatively after removing the nut from the C3 polyaxial screw on the right; a pressure ulcer in the C6 vertebral arch on the left *(2)*

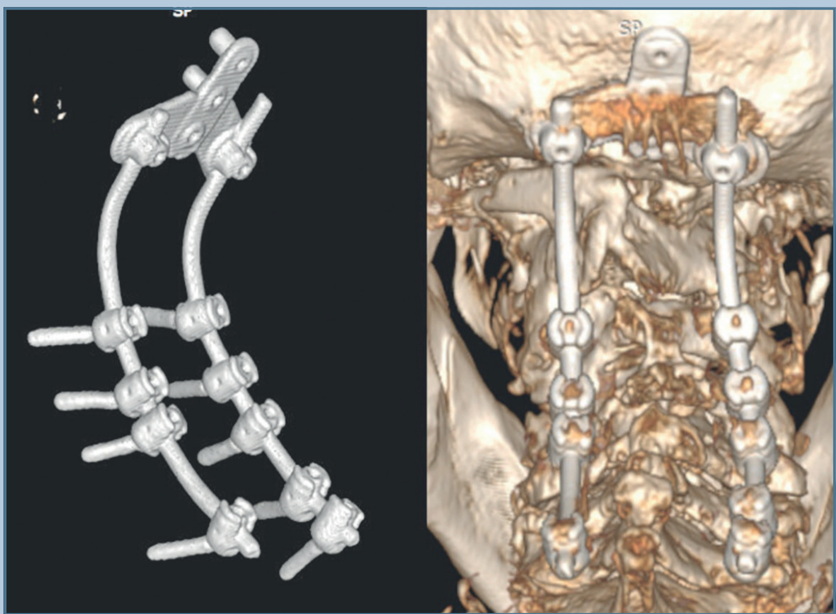

Fig. 6
SCT (3D reconstruction) of patient S. after the 1st stage of surgical treatment; reinstallation of occipitocervical fixation, installation of additional polyaxial screws in the C6 and C7 vertebrae and extension of the parallel rods of the structure to the C7 vertebra with indirect decompression of the spinal cord

Table 3

Parameters of the cervical balance of patient S.

Parameter	Before the second surgery	After the second surgery
C0–C2 angle, degrees	31.8	31.9
C2–C7 angle, degrees	12.7	22.0
T1 slope, degrees	24.6	25.8

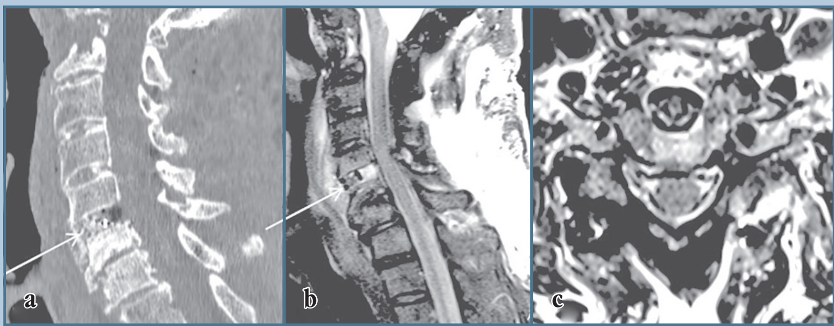

Fig. 7
SCT **(a)** in sagittal, MRI in sagittal **(b)** and axial **(c)** planes after the second stage of surgical treatment: anterior discectomy with decompression of the spinal cord and cage fixation of the C5–C6 vertebrae **(a)**; distal junctional kyphosis is reduced, compression of the spinal cord is eliminated, the size and shape of the focus of the altered MR signal have decreased **(b, c)**

provided decompression of the spinal cord and reliable fixation that resulted in a considerable improvement in the functional independence of the patient.

Eight years after, the onset and aggravation of myelopathy were associated with the progression of distal junctional kyphosis due to a fracture of the rod and dislocation of the OCF system. The patient underwent a two-stage reconstructive and stabilization surgery: partial reinstallation of elements of the OCF system and anterior discectomy with spinal cord decompression and cage fixation in the C5–C6 segment.

Conclusion

The given rare clinical observation demonstrates a combination of basilar invagination with assimilation of the atlas that has determined two-stage surgical treatment in one surgical session (the first stage: decompression; the second stage: OCF). The use of extended systems in occipitocervical fixation should be considered a risk factor for adjacent segment disease development. This observation validates the widely recognized fact that an injury episode is often a trigger for the development of clinical symptoms in patients with abnormal CVJ development in both childhood and adulthood.

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The study was approved by the local ethics committees of the institutions. All authors contributed significantly to the research and preparation of the article, read and approved the final version before publication.

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