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(CASE REPORT)

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Application of "key and lock" principle in the treatment of traumatic cervical Spondyloptosis of c6-7: Case report and literature review

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Abstract

Purpose: Traumatic cervical spondyloptosis is a rare clinical pathology that often causes complete paralysis. When a patient presents neurologically intact or incomplete paralysis, the management becomes more complicated. Preservation of neural function and restoration of anatomic alignment collectively remain the golden goals of therapy. There are still few reports on this issue, however, the existing literature did not clearly define severity of injuries and the treatment strategy in such cases. On the occasion of reporting one treated traumatic C6-7 spondyloptosis case, we reviewed existing literatures.

Material and Methods: The patient was a 48-year-old man who had motorcycle accident and was presented with postoperation for right frontal epidural hematoma in 3 days before admitting to hospital. Neurological assessment showed motor strength grade 3/5 in the proximal upper and lower-extremity muscle groups on the right side, and 1/5 on the left side with incontinence of sphincters. X-rays and computed tomography (CT) scan revealed a three-column ligamentous injury with complete anterior displacement of C6 to C7 vertebral body. We applied the "Key - Lock" principle for better understanding of the biomechanics of the injury and found out the favorable strategy. The patient underwent posterior approach with decompression and anterior approach with reduction and anterior cervical discectomy fusion for C6-7.

Results: After 8 months follow-up, the patient had recovered muscular strength in bilateral upper and lowerextremities muscle groups, sphincter function had fully recovered, and he was able to ambulate by himself. Plain radiograph and CT scan showed good aligment and progressive maturation of his fusion procedure.

Conclusion: Traumatic cervical spondyloptosis of C6-7 is a high-energy unstable fracture and rare. This report develops a new principle called "Key and Lock" for more understand the biomechanics of the injury and treatment. Our patient was successfully treated with open reduction, decompression, and anterior cervical discectomy fusion.

Keywords: Traumatic cervical spondyloptosis; Spine injury; "Key - Lock" principle; Spine injury classification.

1. Introduction

Cervical spondyloptosis is one of the most severe spinal cord injuries or spondylolisthesis, when the entire upper vertebral body is completely slipped to the anterior border of the lower vertebral body [1]. Cervical spondyloptosis is rarely reported, there have some case reports on worldwide. The cause of dislocation can be birth defects, obstetric trauma, accidental trauma, infection, tuberculosis of the spine, or primary tumor. In cervical spine trauma, patients often

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experience severe spinal cord injury, leading to total motor-sensory paralysis and respiratory muscle paralysis. The main goal of the treatment process is to straighten, decompress and stabilize the spine through surgery. In addition, in some known cases the patients had incomplete spinal cord paralysis or even without any neurological damage. At this point, surgery poses a huge challenge to preserve clinical symptoms. Moreover, the limited number of cervical vertebrae, the sensitiveness, and importance of the spinal cord are factors that contribute to great difficulties for the surgery. The treatment strategies reported by authors around the world were often based on individual experiences, thus being uncoordinated as well as controversial. Some authors advocated the use of anesthesia for pre-operative cervical traction, followed by anterior surgery to fuse one or more spinal segments, and finally posterior decompression - fixation surgery of the spine. Other surgeons suggested posterior decompression surgery, then discectomy or corpectomy by anterior approach, then finally the posterior spinal fixation. However, many authors didn't support the pre-operative anaesthesia for dislocation closure for fear of possible nerve damage or vertebral artery injury during operation.

We reported the treatment result of a case of C6-7 cervical spondyloptosis due to trauma, associated with brain injury and incomplete spinal cord paralysis. The "Key - Lock" principle was applied as a key factor to better understand the mechanism of injury and devised reasonable treatment strategies for other cases. To our knowledge, this is the first report of posterior decompression, direct anterior straightening, and single-segment spinal fixation for this rare case of injury.

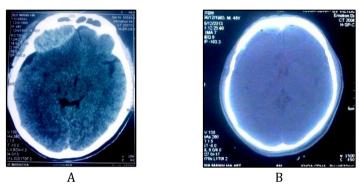


Figure 1 A Preoperative cross-sectional CT scan of traumatic brain injury, demonstrating right frontal epidural hematoma; B. Post-operative cross-sectional CT scan, demonstrating complete resolution of hematoma

2. Case report

2.1. Medical history and clinical symptoms

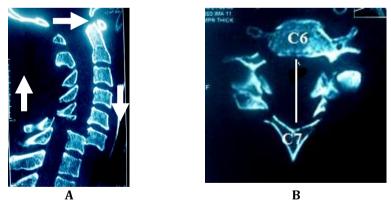


Figure 2 A CT scan showing dislocation of the vertebral body in the forward direction C6C7. The C6 body lies completely in front of the C7 body. The posterior arch and facet joints are broken, preventing compression of the spinal cord from the back. **B**. Cross-sectional CT scan across the dislocation. The spinal canal is enlarged, represented by the white line, the result of vertebral dislocation, complete fracture of the posterior elements.

The case was a 48-year-old male patient who was involved in an accident while driving his motorbike at high speed. He was diagnosed with traumatic brain injury, right frontal epidural hematoma, with a Glasgow Coma Scale (GCS) was 12 points and had undergone hematoma removal surgery to reduce brain pressure at another hospital prior. He was

admitted into Viet Duc University Hospital upon the third day recorded post-operation. Clinical examination upon admission showed that the patient was fully awake, had a GCS score of 15 points, left hemiplegia with left arm, and left leg muscle strength of 1/5. Muscle strength of the right arm and leg was at 3/5. The notable sign was that the patient did not have any sensory disturbances from C6-7 and below. The patient had accompanying bladder dysfunction and required urinary catheterization.

2.2. Medical imaging

On the CT scan of the brain, the pre-operative right frontal epidural hematoma could be seen and the result of craniotomy (Figure 1). CT scan of the cervical spine with longitudinal section demonstrates C6-7 spondyloptosis (Figure 2). The fracture of the C5 joint and the posterior C6 arch helped preventing the bone fragment from entering the spinal canal.

MRI result once again proved the presence of spondyloptosis. Although the spine was completely dislocated, presence of cerebrospinal fluid signals (CFS) in the dorsal part of the spinal canal was evidence for complex fracture of multiple components behind. It was this fracture that causes the spinal canal to widen, preventing spinal cord injury at the time of the accident (Figure 3A).

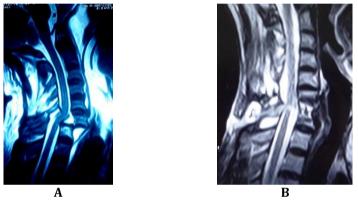


Figure 3 A T2-weighted MR pre-operation. Despite complete dislocation of the vertebral body, cerebrospinal fluid (CSF) was present in the dorsal side of the spinal cord, because of multiple posterior fractures, widening the spinal canal itself. **B**. T2-weighted MRI 6 weeks post-operation. Spinal straightening result were good, the CSF was circulating, but leakage of the cerebrospinal fluid from the point of incision could be observed.

2.3. Classification

According to the classification of subaxial cervical spine injury of Alexander R. Vaccaro, our patient scoring method was as follows: Injury morphology: spondyloptosis = 4 points; Disco-ligamentous complex: complete rupture = 2 points; Neurological status: incomplete spinal cord paralysis = 3 points [2]. Thus, the total score of the patient was 9 points and there was an absolute indication for surgery. However, the patient also had an aggravating factor for the whole physical condition, which was traumatic brain injury and previous craniotomy. We could not be certain of the prognosis and the consequence of this factor for the upcoming surgery. This was the first clinical case with associated traumatic brain injury in the medical literature.

2.4. Methods of surgery

First, the patient was put under general anesthesia and placed in the prone position. We made a midline incision, from C5 to T1. After dissection of the bilateral paravertebral muscle, anatomical injury showed complete rupture of the posterior ligament due to tension, complete fracture of the posterior arch C6 and part of C5, the left C6-7 joint surface was fragmented, entering the conjugate hole. The patient underwent a C5C6 lamina removal, extensive decompression, release of many extramedullary hematomas, and complete resection of the bilateral C6-7 joints. Cerebrospinal fluid overflowing was also found, however, no posterior dural tear was found. The cause of this could be a tear in the lateral side or front of the spinal cord, which could not be detected. Due to the patient's severe condition and prolonged double-section surgery, we decided to cover the dura mater with spongel and close the fascia meticulously with each anatomical layer.

Next, the patient was turned over in the supine position. A percutaneous incision was made at the level of the left C6-7 disc. according to the technique of Smith-Robinson. When dissecting to the anterior longitudinal ligament, the soft tissue

was edematous, spreading around the C6 body protruded anteriorly. After ligamentectomy, the C6-7 disc completely ruptured and partially dislocated anteriorly. We removed most of the disc to create a space between the two vertebral bodies. Then, we put the splint expander into the space just created, slowly stretch the two vertebral bodies. When the stretching distance was large enough, we increased the pressing pressure on C6 vertebrae from front to back to achieve absolute straightening. After that, the surgeon continued to remove the remaining disc and discovered that the posterior longitudinal ligament was completely tear, intervertebral disc fragments were removed from the back of the C6 vertebral body. Up to this point, we still did not detect any cerebrospinal fluid leak or dura tear. After preparing the graft, a piece of bone from the patient's left iliac wing was used to replace the removed disc. Finally, we placed an anterior neck brace 26 mm long and secured 4 screws 14 mm long into the C6-7 vertebral body. The continuous drainage was put before the anatomical layer closure of the muscle fascia.

2.5. Follow-ups

The postoperative CT scans demonstrated very good straightening results (Figure 4). The patient's neurological symptoms improved immediately post-operation. Then patient was transferred to the rehabilitation center after 6 days of surgery, with a stiff cervical collar. Six weeks later, the patient came back for a follow-up examination with good progress: complete restoration of hand and leg motor function, normal walking, and good bladder function. However, there was leakage at the end of the 1cm long incision. Post-operative MRI showed a leak of cerebrospinal fluid outside the skin (Figure 3B). However, the patient was still treated conservatively and took prophylactic antibiotics. Two weeks later, the fistula healed well and no complication was recorded. Eight months after surgery, radiographs reflect very good bone healing results.



Figure 4 Postoperative CT scan demonstrates good orthopedic results, with anatomical structure restored to normal. A. Sagittal CT scan. B. Coronal CT scan

3. Discussion

In 1882, Neugebaner first described the concept of spondyloptosis, which is a term for more than 100% dislocation of the vertebral body at any position in the spine. In 1951, Perlman and Hawes [3] reported the world's first clinical case of cervical spondyloptosis. Since then, some articles on the topic of cervical spondyloptosis had been published on the PubMed and Ovid databases, regarding the location of the lesion, the trauma factors, and the treatment [4]. In trauma, cervical spondyloptosis usually occurs because of a head-on fall while riding a motorcycle at high speed or falling headfirst into water. Our patient, the primary mechanism causing the injury was due to axial compression force and excessive spinal extension after a motorcycle accident (note the right frontal epidural hematoma). When the cervical spine was overextended and subjected to a compressive force due to the head hitting a hard object, the traumatic force impacted directly on the posterior spine, causing a complete fracture of the lamina on both sides and rupture of the facet joints and lateral masses. Next, the excessive force of the spine completely tore the anterior longitudinal ligament, the intervertebral disc, and the posterior longitudinal ligament of the C6-7 vertebrae. By inertia, the entire body of the C6 vertebra moved completely in front of C7, causing the phenomenon of cervical spondyloptosis, or in other words, the C6 vertebra was locked in front of C7. It was interesting that besides human trauma, nature provided an excellent selfdefense mechanism by expanding the spinal canal. It was the complete fracture of the posterior elements of the spine that caused the spinal canal to become wider, or in other words, to self-decompress, to help prevent bone fragments from pressing on to the spinal cord. This phenomenon explained why the patients had only temporary spinal cord injury, incomplete paralysis, or even no neurological deficit. Indeed, Merianos observed 17 cases of cervical spondyloptosis with bilateral pedicle fractures. The author emphasizes that there were 13 cases without neurological damage [5] and this was also consistent with our patient who was incompletely paralyzed.

Stemming from observations in real life, we found that operating a lock required the key to be inserted into the cylinder and turned clockwise. This was similar in case of cervical spondyloptosis, where it was the axial compression and spinal overextension that initiate the key insertion, causing the C6 vertebral body to move clockwise and lock in front of C7 (Figure 2A). We called this phenomenon "direct dislocation" and developed the principle of "Key-Lock" dislocation. To put it back, if the lock was inserted upside down, what would happen? According to the principle above, to initiate the lock we now need to turn the key counterclockwise. Indeed, while it was very rare, there were cases of cervical spine injury, where the superior vertebrae moved counterclockwise and bloked behind the inferior vertebrae (Figure 5), which was called "inverted dislocation". The injury mechanism by then came from the compression force. Under the pressure of excessive bending force and axial compression, the lower border of the upper vertebral body would rupture, tearing the ligament-disc complex, and as a result, the upper vertebral body completely dislocated posteriorly to the inferior vertebral body. Thus, according to the "Key-Lock" principle, in cervical spine injury, there will be two types of cervical spondyloptosis, direct and inverted dislocation, with completely different locking of the vertebral body. So, is the unlocking method of these two types of dislocation also not the same?

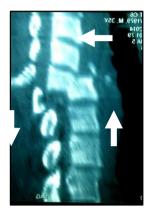


Figure 5 Sagittal CT scan of longitudinal section, demonstrating inverted dislocation. The superior vertebral body ruptured, shifted counterclockwise, dislocating posteriorly to the inferior vertebral body.

For cervical spondyloptosis patients with incomplete or even non-paraplegic cervical spondylosis, straightening surgery presents a tremendous challenge. The first and foremost goal is to protect nerve functions from further damage. Along with that, a supreme task is to surgically straighten the dislocation, decompress and stabilize the spine, thereby prevent secondary nerve damage, facilitate the restoration of nerve function, and avoiding complications caused by spinal instability. Nowadays, there were some reports about the different surgical techniques for such type of injury. In 2009, Tumialan et al. [6] reported a case of C7-T1 traumatic spondyloptosis without neurological damage. The patient was anesthetized for close straightening and fixation on one level of C7-T1 from the front, then fixed with the C6-T2 screw from the back during the same surgery. Bhojras et al observed an 8-year-old girl with dislocated C6C7 vertebral bodies after obstetric trauma, presenting with delayed nerve compression. The pediatric patient was treated with anterior C5C6C7 vertebral ablation, autologous pelvic transplantation [7]. Shah [8] reported a case of traumatic C6-7 spondyloptosis, which was successfully treated with anterior tract surgery without instrumentation. In 2014, Kumar Ravi et al. [9] performed surgery on a patient with traumatic spondyloptosis C5-6, the patient was still able to walk. The mentioned author successfully treated C6 corpectomy, bone grafting with titanium cage, and fixed by simple screws. On the contrary, many authors opposed the use of pre-anesthesia traction before surgery, given the evidence of neurological complications during straightening. Mahale et al. [10] reported 16 patients with nerve damage after cervical spine straightening, including 4 cases with closed pre-anesthesia traction. The reports found in the worldwide database on treatment strategies for cervical spondyloptosis were often based on personal experience, inconsistent, and rather controversial. The key point was that mentioned authors did not clearly classified the type of spondyloptosis. did not generalized the mechanism of injury of each type, form which the corresponding straightening principle could not be defined. Therefore, spine surgeons often faced great challenges in choosing the proper surgical techniques.

In our case, the patient was presented with incomplete spinal cord paralysis and the right frontal epidural hematoma that was previously removed. At this time, the surgery faced many challenges when it came to preserving neurological functions and poor prognostic factors caused by traumatic brain injury. Moreover, we did not have a standard preoperative anesthesia closed cervical traction system, along with the inconsistency in surgical techniques in the literature, which had caused considerable confusion in the treatment strategy. To answer those questions, we looked back to the "Key - Lock" principle. Indeed, to open a lock we simply needed to turn the key counterclockwise. Like

patients with direct spondyloptosis, what needed to be done was to reverse the C6 vertebral body and, in other words, to reverse the process of spinal locking. Therefore, we extended 2 vertebrae C6-C7 with a splint to relieve axial compression. Then, applied pressure on C6 vertebrae from front to back to reverse the displacement caused by overextension of the spine and result in successful dislocation. The initial posterior decompression surgery was not intended to secure the spinal cord during straightening and to facilitate the decompression process by removing the bone fragments trapped in the middle. It could be said that, with the reverse technique of spinal locking, based on the "Key - Lock" principle, the author had overcome the limitation due to the lack of a preoperative anesthetic closed cervical traction system, successfully straightening the dislocation intra-operative, safely preserve neural function. Besides, we had rationalized the treatment strategy compared with other authors. So, the follow-up question was if we could use the above straightening technique to unlock the inverted dislocations (Figure 5). The answer was no. We could not increase the pressure on the lower vertebrae from front to back to achieve balance with the upper vertebrae, while the upper vertebra was pressing on the spinal cord. It would be a disaster caused by the surgeon. To answer this question, we still applied the principle of "Key - Lock". Also, we needed to turn the vertebral key clockwise. The goal of surgery was to reverse the process of spinal locking, eliminating the traumatic forces. Therefore, the surgery needed to have both anterior and posterior approaches. Via the anterior passage, although the lower vertebra was not ruptured, it was necessary to remove it to open the entrance to the upper vertebra that was compressing the spinal cord. The upper vertebrae must also be amputated to eliminate the compression and over bend of the spine, and then completely decompress the spinal cord. Next, the patient needed a bone graft and a neck brace first. Finally, posterior surgery to fix the spine through the dislocated segment was needed to ensure 360° stability. Based on our knowledge, we believed that this was the first report on the straightening technique based on the "Key-Lock" principle. The application of this principle would help spine surgeons to better understand the mechanism, classify the injuries, and devised reasonable treatment strategies for many similar cases.

One noteworthy thing was the posteriorly herniation of the C6 vertebral body. In the process of vertebral dislocation, the posterior longitudinal ligament would be stretched along the posterior border of the upper vertebrae, even breaking completely. At this time, the anterior border of the spinal cord would be extended upward, large disc fragments could herniate to the posterior border of the C6 vertebral body, without compressing the spinal cord due to the newly created space. However, after the process of straightening, the herniated fragment would cause secondary compression due to the absence of an anterior space. Therefore, it was necessary to remove all the migratory hernia fragments before performing bone grafting. The author also found that the careful examination of bone lesions on preoperative CT scan and meticulous assessment of intraoperative anatomical lesions helped make the most accurate final decisions. In cases of severe spinal injury, fracture of the entire posterior complex, especially if the vertebral column on both sides was completely broken, the author recommended that it was mandatory to combine posterior surgery to fix the spine, ensuring the 360° configuration. With our patient, although we did not detect any dura tear causing CSF leakage during the surgery. However, the patient still appeared to leak cerebrospinal fluid through the skin incision post-operative. It may be due to undetectable lateral or anterior lacerations of the spinal cord. Therefore, a thorough, multi-layered incision closure and placement of a drainage tube to reduce CSF pressure in the lumbar region will help prevent post-operative fluid leakage, as well as meningitis complications.

4. Conclusion

Cervical spine injury C6-7 spondyloptosis is a complex and very rare injury. Surgical manipulation for patients without nerve damage or incomplete spinal cord paralysis is a huge challenge. Mastering the "Key-Lock" principle will help to better understand the mechanism of injury and devise an appropriate treatment strategy. The application of the above principles had helped the author successfully treat with reverse locking technique and fix the spine one level first.

Compliance with ethical standards

Acknowledgments

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Disclosure of conflict of interest

The authors declare that they have no conflict of interest.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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