Revision of Adjacent Spondylopathy after Cervical Occipital Fusion: A Case Report

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1. Abstract
1.1. Purpose: To share our experience and lesson in treating 1 case of revision of adjacent spondylopathy after cervical occipital fusion. Surgical treatment is widely adopted in the adjacent spondylopathy after cervical occipital fusion, but what kind of surgical treatment is rarely discussed.

1.2. Methods: One patient underwent anterior cervical spine surgery because of adjacent spondylopathy after cervical occipital fusion. Implant failure was not observed intraoperatively and occurred on postoperative day 2. Afterwards, second revision anterior cervical spine surgery was carried out immediately. Ten days after the second revision surgery, the screw loosened again, and third revision cervical spine surgery via a combined anterior and posterior approach was performed.

1.3. Result: At 15 months after the third revision surgery, the patient recovered well without complications or screw loosening.

1.4. Conclusion: Surgical repair succeeded in this patient who underwent adjacent spondylopathy after cervical occipital fusion.

2. Introduction
Posterior Occipito-cervical Fusion (OCF) is an effective and reliable surgical procedure for the treatment of skull base depression and occipito-cervical junction deformity, and has achieved good clinical outcomes[1]. However, there is almost no extension, flexion and rotation in the upper cervical spine after OCF, leading to limited range of motion in the neck [2, 3]. After OCF, most neck motion is mainly concentrated in the lower cervical spine. Stress concentration at the region of the occipito-cervical junction may result in the development of Adjacent Segment Disease (ASD) and also negatively influences clinical outcomes, then revision surgery is often needed. Due to the stress changes, the risk of failure of revision surgery increases. Improper procedure selection during revision surgery may cause serious consequences in patients. Herein, we reported a case of ADS following OCF who was admitted to our hospital on July 2019, and underwent multiple revision surgeries for implant loosening after anterior cervical discectomy (decompression) and fusion (ACDF) for ASD.

3. Case Report
A 46-year-old female patient was admitted to our hospital due to numbness of extremities and unsteady walking for 1 year. She had undergone OCF for skull base depression in the local hospital, and showed fair recovery. Neurologic examination showed that the strength of intrinsic muscles of both hands and key muscles of both lower extremities were graded as IV, there were decreased tactile sensation, normal pain sensation in the left hand and both lower extremities, as well as normal sensation in the saddle region. She had a Japanese Orthopaedic Association (JOA) score of 11. Anteroposterior and lateral X-ray and CT scanning of the cervical spine showed cervical spine instability (Figure 1 A-F). MRI of the cervical spine revealed cervical disc herniation at C3/4 level and cervical spinal cord compression (Figure 1G).

Preoperative planned surgical procedure for the patient was C3/4 ACDF, combining bone graft with anterior plate fixation. During surgery (on July 16), we found that cervical spine extension in the patient was obviously restricted, so the desired decompression effect cannot be achieved by using the planned procedure. The surgical procedure was then altered to C4 anterior cervical corpectomy and fusion (ACCF), combining implantation of the artificial vertebral body with anterior plate internal fixation. Two days after the first surgery (on July 18), the patient complained of dysphagia after strenuous extension exercises of the head and neck. Anteroposterior and lateral X-ray of cervical spine showed that the anterior cervical plate loosened, tilted out and squeezed the esophagus (Figure 2A-B). Considering that dysphagia may be
further aggravated and even severe complications such as esophageal fistula and asphyxia may be caused, an emergency revision anterior cervical spine surgery was performed.

On July 23, anteroposterior and lateral X-ray of the cervical spine after the second revision surgery documented good position of the anterior cervical plate. Ten days after the second surgery (on July 28), the patient complained of dysphagia again. Cervical spine x-ray re-examination showed that the cervical spine plate loosened and the screws pulled out (Figure 2 C-D), then revision cervical spine surgery with a combined posterior and anterior approach was performed on an emergency basis. During the third surgery, the screws, plates and artificial vertebrae were first removed via the anterior approach, then the incision was temporarily sutured. Afterwards, lateral mass screws were placed bilaterally at C3, C4, and C5 through the posterior approach and secured with connecting rods along with the screws placed during the primary OCF. Finally, the trimmed iliac bone blocks were implanted in the anterior sulcus through the anterior approach

During the early period after the third revision surgery, relief of dysphagia was seen in the patient. After 2 weeks of surgery, movement and sensation in all four limbs of the patient were obviously improved compared with before surgery, the patient was then discharged. Neurologic examination showed that the strength of key muscles of four extremities was graded as V. The JOA score was 13. After 15 months of surgery, reconstructed CT sagittal scan showed good autogenous bone graft fusion (Figure 2G). After rehabilitation treatment, the patient showed varying degrees of functional improvements in all four extremities, confirming the effectiveness of the revision surgery.

**Figure 1 a, b, c, d:** Preoperative extension and flexion X-ray show C4 instability  
**e, f:** Preoperative CTs sagittal position show C4 instability  
**g:** C3/4 disc herniation, and cervical spinal cord compression.

**Figure 2 a, b:** X-ray of neck at two days after the first surgery showing plate slip  
**c, d:** X-ray of neck at ten days after the second surgery showing screw out  
**e, f:** X-ray of neck at one week after the third surgery showing good internal fixation position  
**g:** CT of neck at 15-month follow-up after the third surgery showing good bone graft fusion  
**h:** MRI of neck at 15-month follow-up showing normal signal of cervical spine.
4. Discussion

The atlantoaxial joints are primarily responsible for the rotation of the cervical spine, and the C0/C1 segment contributes 23 to 25 degrees of the flexion and extension of the skull [4]. Fusion of the atlantoaxial joints greatly influences the cervical flexion and extension motions. Compared with fusion of the atlantoaxial joints alone, OCF results in more significant limitation of motion. The lower cervical spine compensates for the motion unit by compensating itself, leading to corresponding increase in the range of motion, this may be an important reason for changes in the curvature or even accelerated degenerative changes of the lower cervical spine [5]. Biomechanical studies[6, 7] have shown that after OCF or atlantoaxial fusion, the increased stress on the lower cervical accelerated degeneration, leading to the occurrence of ASD, and severe cases may require reoperation.

ACDF is less traumatic, easy to operate, and causes less damage to normal anatomical structures, which is not only the main surgical procedure for primary treatment of cervical spine degeneration, but also the surgical procedure widely used in revision cervical spine surgery. When choosing surgical procedure for revision cervical spine surgery, it is important to consider not only the effect of decompression, but also the maintenance of strength after fixation, as well as the influence of stress changes on flexibility of the cervical spine, inadequate consideration may lead to the failure of the revision surgery and cause serious consequences. In this study, the patient developed ASD after OCF, there are more pitfalls in selecting surgical procedures for reoperation in this patient, anterior fixation was selected as the first revision surgery for ASD, and due to the restricted cervical spine extension after OCF, it is difficult to perform anterior decompression with screw fixation during reoperation. The preoperatively planned surgical procedure was single-segment ACDF, which was changed to ACCF during surgery because there was no enough space for adequate decompression, this set the stage for failure of the surgery.

Both anterior and posterior cervical spine surgery can achieve good results, especially the anterior cervical spine surgery, which can relieve the pressure directly and exhibit definitive decompression effect. However, for patients who developed ASD after OCF, although a good decompression effect can be achieved when the anterior approach is chosen, but the continuity of the biological force line is interrupted after fixation. When the stress is concentrated at the junction of anterior and posterior internal fixation, especially when the force arm is increased after corpectomy, screw stress at the junction area is higher and the risk of surgical failure is obviously increased. In this case, during the second surgery, although we found that the inferior endplate of the C3 vertebral body was damaged, the superior endplate has sufficient bone density to provide initial fixation strength, but we still did not pay attention to the mechanical continuity after fixation, chose to lengthen the fusion titanium cage, continued to perform titanium plate fixation via the anterior approach, and did not perform extended posterior fixation, resulting in the failure of the second revision surgery. Although the initial stability immediately after revision anterior cervical spine surgery was good, but these two revision surgeries ended in failure due to stress changes, and good stabilization results were not obtained even with the use of external fixation brace. In clinical practice, attention should be paid not only to immediate stability, but also to continuous stability before bone fusion. External fixation with neck brace cannot provide the continuous stability required for bone fusion.

Screw pullout poses increased risk of tracheal or esophageal damage. Esophageal fistula after anterior cervical spine surgery is one of the rare but serious complications [8, 9]. Yee et al. [10] reported a case of esophageal perforation caused by screw pullout, and the screw was expelled via the digestive tract, the patient had no relevant clinical symptoms. Nevertheless, we believe that if there is clear imaging evidence confirming screw pullout, immediate revision surgery or removal of internal fixation should be performed in order to avoid secondary damage to important tissues or organs.

In this study, although previous two revision surgeries dealt psychological blows to the patient, the patient agreed to receive third revision without hesitation. Considering that there is no enough space for plate fixation via the anterior approach, so the surgical plan was as follows: the internal fixation was removed via the anterior approach, possible factors contributing to compression were eliminated, then the incision was temporarily sutured, patient’s position was changed, the fixed segment via posterior approach was extended to the C5 vertebral body, patient’s position was changed again, then iliac bone was harvested and grafted. A head-neck-chest brace was used for external fixation after surgery, good bone fusion was achieved after 3 months of surgery.

In the present case, the sticking point in revision surgery is surgical approach selection. Implant loosening was observed in the patient after two previous revisions, suggesting that stress factors may be central to the success or failure of the fusion surgery, better fixation can be obtained through an extended posterior approach. Lateral mass screw fixation and posterior pedicle screw fixation are the most commonly used and effective technique in posterior cervical spine surgery, which have good biomechanical properties, can stabilize the cervical spine and reconstruct cervical spine physiological curvature [11, 12]. Because during the two previous revisions, the vertebral body was resected via the anterior approach, so when performing posterior cervical spine fixation, autologous bone grafts should be fully used to increase the bone graft fusion rates. The quality and shape of the block bone graft have an important influence on the stability of the entire fixed segment. In this case, autologous tricortical iliac bone block were used. During the surgery, attention should also be paid to increase the contact area between the bone grafts and the bone bed, i.e., bone grafting should be performed adequately and effectively in the end-to-end...
and side-to-side fashion, in order to achieve firm bone fusion. In addition, it is important to understand the patient’s psychological state, work environments and any compensation claims for medical disputes that may arise. In this study, the patient was finally successfully treated after multiple revisions, this is inextricably linked with the understanding and cooperation of the patient and her family members.

Revision surgery for the treatment of ASD after OCF should be performed following the principles of individualization. Patients’ age, etiology, primary surgical procedures that patients received and the impact of stress changes on reoperation need to be fully considered. In addition, sufficient preoperative evaluation and selection of the appropriate surgical approaches and methods of fixation are required, and direct decompression should not be excessively pursued. The revision surgery requires complete relief of the existing compression and restoration of biomechanical stability as much as possible in order to maintain a stable mechanical environment for the enhancement of bone fusion, thereby reducing the risk of surgical failure.

5. Conclusion

Revision surgery for the treatment of ASD after OCF should be performed following the principles of individualization. Patients’ age, etiology, primary surgical procedures that patients received and the impact of stress changes on reoperation need to be fully considered. In addition, sufficient preoperative evaluation and selection of the appropriate surgical approaches and methods of fixation are required, and direct decompression should not be excessively pursued. The revision surgery requires complete relief of the existing compression and restoration of biomechanical stability as much as possible in order to maintain a stable mechanical environment for the enhancement of bone fusion, thereby reducing the risk of surgical failure.

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