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ORIGINAL ARTICLE

Anterior controllable antedisplacement fusion as a choice for 28 patients of cervical ossification of the posterior longitudinal ligament with dura ossification: the risk of cerebrospinal fluid leakage compared with anterior cervical corpectomy and fusion

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Abstract

Purpose To compare the incidence rate of cerebrospinal fluid (CSF) leakage between anterior controllable antedisplacement fusion (ACAF) and anterior cervical corpectomy and fusion (ACCF) in the treatment of ossification of the posterior longitudinal ligament (OPLL) with dura ossification (DO).

Methods In the period from June 2015 to June 2017, ACAF and ACCF were performed on patients with OPLL with DO. Double-layer sign was observed on axial bone window of CT images. The operation duration, blood loss, and hospital stay were measured. Radiologic assessment included occupying rate, type and extent of OPLL, decompression width, postoperative area of the spinal canal, and anteroposterior diameter of the spinal cord. The JOA scoring system was used to evaluate the neurological status. Surgery-related complications such as CSF leakage and spinal cord or nerve injury were all recorded.

Results There were 28 patients in ACAF group and 31 in ACCF group. On cross-sectional CT, decompression width and postoperative spinal canal area were both significantly larger in the ACAF group than that in the ACCF group ($P < 0.01$). The anteroposterior diameter of the spinal cord was significantly larger in the ACAF group ($P < 0.05$). Mean JOA score was better in the ACAF group ($P < 0.05$). In the ACCF group, seven (22.6%) patients had CSF leakage. However, only one (3.6%) presented with CSF leakage in the ACAF group. The difference of incidence rate of CSF leakage was significant ($P < 0.01$).

Conclusions ACAF, which can significantly reduce CSF leakage and achieve good neurological recovery, is a good option to treat cervical OPLL with DO.

Haisong Yang and Jingchuan Sun contributed equally to this paper.

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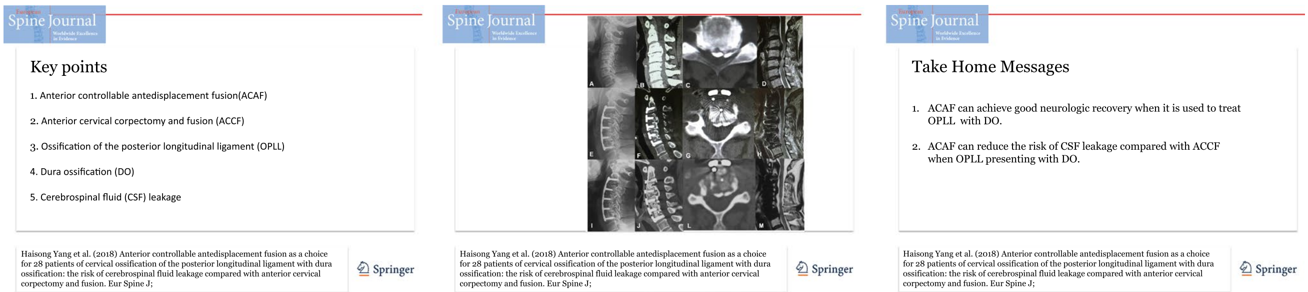
Extended author information available on the last page of the article

Graphical abstract These slides can be retrieved under Electronic Supplementary Material.



Keywords Ossification of the posterior longitudinal ligament (OPLL) · Anterior controllable antedisplacement fusion (ACAF) · Anterior corpectomy and fusion (ACCF) · Dural ossification (DO) · Cerebrospinal fluid leakage · Double-layer sign

Introduction

Ossification of the posterior longitudinal ligament (OPLL) in the cervical spine often presents with dura ossification (DO). Once this dural membrane is ossified in the association with OPLL, it becomes a technical challenge to separate the ligament from DO [1, 2]. Accordingly, CSF leakage through the defect portion of the dura mater is a common complication encountered when using the anterior approach for direct removal of cervical OPLL [3, 4]. For OPLL with DO, the incidence of CSF leakage is much higher, ranging from 4.3 to 32% [5]. Many authors reported that the presence of DO was the greatest risk factor for developing a CSF leak in the anterior decompression surgery [6–9]. Patients with this diagnosis were 13.7 times more likely to have a cerebrospinal fluid leak during cervical spine surgery [10]. CSF leakage led to a long hospital stay, prolonged recovery duration, high economic cost, increased infection probability, and potential revision surgery [3, 9, 10]. Various secondary complications associated with CSF leakage after cervical operations, including meningitis, delayed wound healing, airway obstruction, cutaneous CSF fistula, and pseudomeningocele, also appeared [11–13]. However, there is still no effective method to prevent CSF leakage.

We have previously presented an anterior surgical technique called "anterior controllable antedisplacement fusion (ACAF)" to treat OPLL [14]. Instead of resecting the OPLL, ACAF moves the vertebrae with OPLL and DO ventrally in a hoisting manner to achieve direct decompression. Here, we compare the incidence rate of CSF leakage and clinical outcomes between ACAF and anterior cervical corpectomy and fusion (ACCF).

Materials and methods

Patient population

The medical records of 28 patients with symptoms of cervical myelopathy who underwent ACAF (17 males, 11 females; mean age 58.0 ± 9.9 years [range, 38–74 years]) and 31 patients who underwent ACCF (21 males, 10 females; mean age 58.7 ± 7.9 years [range, 43–73 years]) from June 2015 through June 2017 at our hospital were retrospectively reviewed. Inclusion criteria were (1) cervical myelopathy, (2) any level of OPLL from C2/3 to C6/7, and (3) dura ossification. Exclusion criteria were (1) spinal trauma, (2) infection, and (3) no double-layer sign on axial bone window of CT images. Indications for surgery were neurological vulnerability due to myelopathy, radiculopathy, and/or intractable pain. Table 1 shows patient demographics and clinical data for both groups.

The SKYLINE® Anterior Cervical Plate System with cage and titanium mesh (DePuy Synthes Spine, Raynham, MA, USA) was used in all patients. Neurophysiologic monitoring including somatosensory-evoked potentials, spontaneous electromyogram, and/or motor-evoked potential intraoperatively was used in all patients. The ethical review board of our institution granted approval to the conduction of this study.

Radiological evaluation

All patients underwent pre- and postoperative X-ray, computed tomography (CT), and magnetic resonance imaging (MRI) of the cervical spine. A diagnosis of OPLL was established for all the patients according to the results of a preoperative radiological examination. Based on the sagittal

Table 1 Demographic, radiological, and clinical outcomes of the two groups

	ACAF	ACCF	*P* value
No. of patients	28	31	
Gender			> 0.05
Male	17	21	
Female	11	10	
Age (year)	58.0 ± 9.9	58.7 ± 7.9	> 0.05
Extent of OPLL			> 0.05
1 level	2	5	
2 level	10	19	
3 level	14	7	
4 level	2	0	
Type of OPLL			> 0.05
Segmental	10	13	
Continuous	7	10	
Mixed	11	8	
Type of double-layer sign			> 0.05
Type A	9	16	
Type B	10	11	
Type C	9	4	
Occupying rate (%)	54.2 ± 7.7	51.5 ± 11.2	> 0.05
Operation duration (minute)	155.2 ± 40.6	161.6 ± 35.2	> 0.05
Blood loss (ml)	289 ± 56	304 ± 81	> 0.05
Hospital stay (day)	6.4 ± 1.1	9.8 ± 7.3	< 0.05
Decompression width (mm)	18.0 ± 1.1	15.2 ± 0.9	< 0.01
Spinal canal area (mm^2)			
Preoperation	77.0 ± 17.9	79.9 ± 20.4	> 0.05
Postoperation	172.5 ± 31.7	140.7 ± 26.9	< 0.01
Diameter of spinal cord (mm)			
Preoperation	2.4 ± 0.8	2.5 ± 1.1	> 0.05
Postoperation	5.5 ± 0.6	5.0 ± 1.1	< 0.05
JOA scores			
Preoperation	8.9 ± 1.2	9.1 ± 1.4	> 0.05
Postoperation*	15.5 ± 0.9	14.7 ± 1.2	< 0.01
Improvement rate (%)	82.4 ± 8.8	71.9 ± 12.4	< 0.05
Complications			
CSF leakage	1	7	< 0.01
SCI or nerve injury	0	0	
Wound infection	0	0	

ACAF anterior controllable antedisplacement fusion, *ACCF* anterior cervical corpectomy and fusion, *CSF* cerebrospinal fluid, *JOA* Japanese orthopaedic association, *SCI* spinal cord injury

*Six months postoperation

image, OPLL was classified into three types: segmental, continuous, or mixed, according to a previous classification [15]. The double-layer sign, characterized by anterior and posterior rims of hyperdense ossification separated by a central hypodense mass, was observed on axial bone window of CT images. Patients with the double-layer sign were diagnosed as DO. There were three types of double-layer sign according to our previous study [9]. The occupying rate (OR) was defined as the thickness of OPLL with DO divided by the anteroposterior (AP) diameter of the bony spinal canal on the axial CT image. Decompression width was the distance between the bilateral grooves at the posterior vertebra. The area of the spinal canal and AP diameter of the spinal cord were also calculated on cross-sectional CT and MRI images, respectively (Fig. 1).

Clinical evaluations

The Japanese Orthopaedic Association (JOA) scoring system was used to evaluate the neurological status before and after the operation. An improvement rate (IR) was calculated as: IR = (postoperative JOA score − preoperative JOA score)/(17 − preoperative JOA score) × 100%. Complications of CSF leakage, spinal cord and nerve root injury, and wound infection were also recorded for the two groups.

Surgical technique

Under general anesthesia, the patients were placed in the supine position with the neck slightly extended. The cervical spine was exposed through a standard right-side anterior approach. The appropriate surgical level was confirmed using intraoperative radiography.

ACCF procedure

After necessary discectomies performed at the appropriate surgical intervertebral spaces, the vertebral bodies were partially removed using an appropriate rongeur. The residual vertebral bodies and OPLL were removed using high-speed drill until the OPLL was thinned as much as possible. Then we tried to separate OPLL from DO through a thin layer consisting of a nonossified degenerated PLL between the OPLL and the DO. This technique has been detailed by Mizuno et al. and Chen, and the ossified portion of the dural mater was carefully preserved to avoid dural defect and CSF leakage [2, 4]. However, the OPLL was not always able to be resected from the ossified portion of dura mater, because the nonossified layer of the PLL was not found in those patients. In this condition, two surgical tactics could be selected. One was the floating method, in which the OPLL was only separated from the around vertebral wall, but remained together with DO. The entire ossified mass could gradually float anteriorly and did not compress the spinal cord eventually. Nevertheless, considering the decompressive effect by the floating method was limited or got very slowly at least, the entire ossified mass including OPLL and DO was removed completely if they could not be separated. This technique needs to be operated with the aid of microscope, and the arachnoid membrane was preserved to avoid a large area of

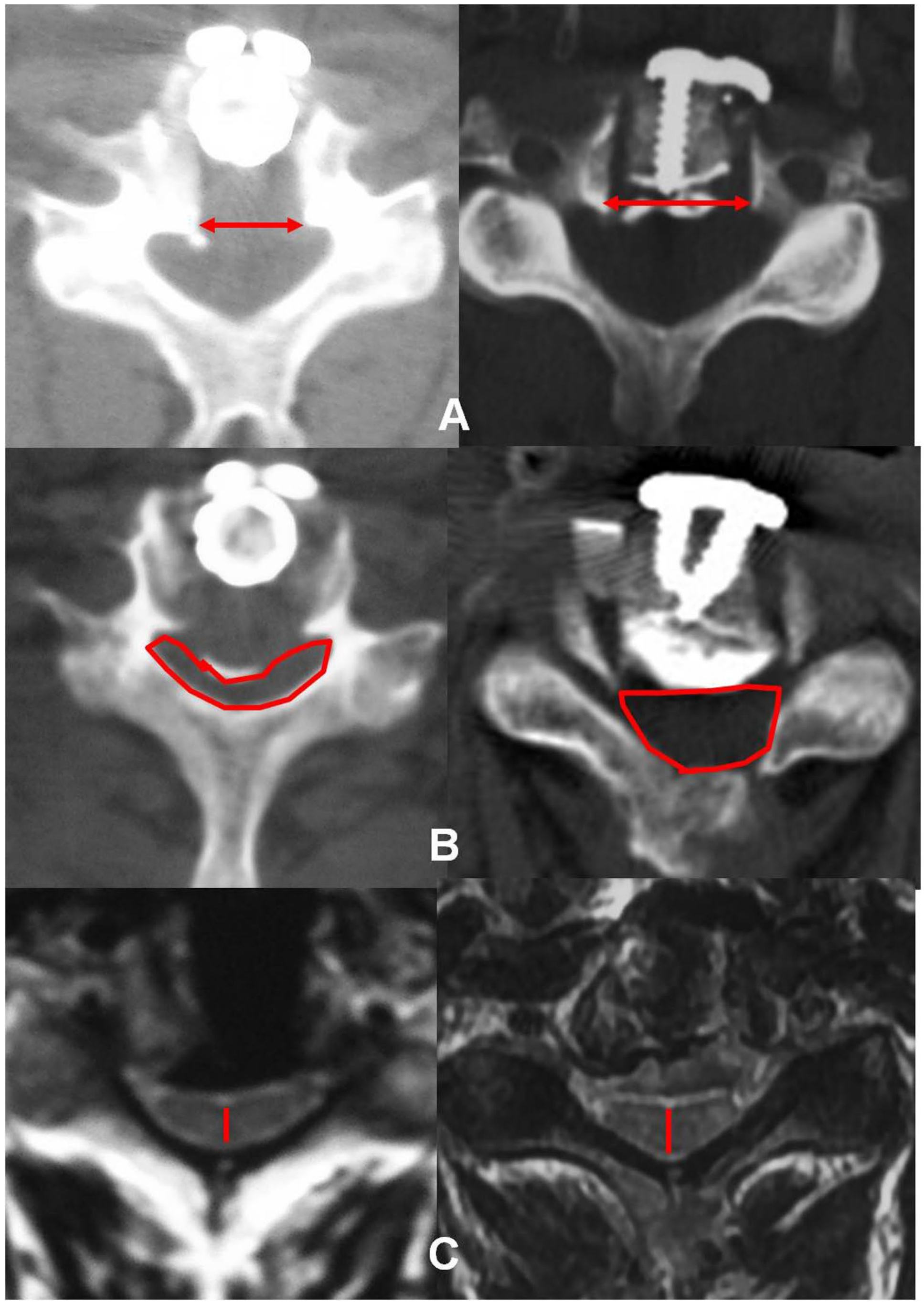


Fig. 1 Measurement of decompression width, spinal canal area, and anterior–posterior diameter of the spinal cord in ACCF group (**a**–**c**, left) and ACAF group (**a**–**c**, right)

membrane defect. After decompression, the titanium mesh cage filled with autologous bone fragments or iliac crest strut together with anterior cervical plate was used to restore the stability of the involved segments.

ACAF procedure

After necessary discectomies performed at the appropriate surgical intervertebral spaces, the posterior longitudinal ligament was cut at the disk spaces corresponding to the caudal and cephalad ends of the ossification and cord compression site. Part of the anterior bone of the vertebrae was then cut according to the thickness of the OPLL and DO. Following this, on the left side of the vertebra, we used a high-speed drill to create a groove about 3 mm wide, approximately at the medial border of the transverse foramina (just at the nutrient foramen of the vertebral bodies), until we reached the posterior wall of the vertebra. This process was repeated at each surgical level. A suitable small cage (4 mm high) filled with autologous bone fragments was then placed into each intervertebral space, and an appropriate over-contoured plate was fixed to the caudal and cephalad vertebrae. On the middle vertebrae, 14-mm-long self-tapping screws were inserted halfway for temporary fixation. On the right side of the vertebrae, as had been performed on the left side, at each surgical level, we created a groove until the posterior wall was reached. Now the vertebrae, ossified ligaments, and DO were isolated. Finally, we tightened the screws in the middle vertebrae, forcing the vertebrae, ossified mass, and DO to move forward in a hoisting manner. Allogenic iliac bone was implanted into the groove to ensure fusion (Fig. 2).

Statistical analysis

Results are presented as mean ± standard deviation. Statistical analyses were performed using SPSS for Windows, Version 12.0 (SPSS Inc., Chicago, IL, USA). The Mann–Whitney *U* test and Chi-square test were used to compare clinical and radiologic outcomes between groups, and Student's *t* test was used to compare differences between preoperative and postoperative clinical and radiologic outcomes. Differences with a *P* value of less than 0.05 were considered significant.

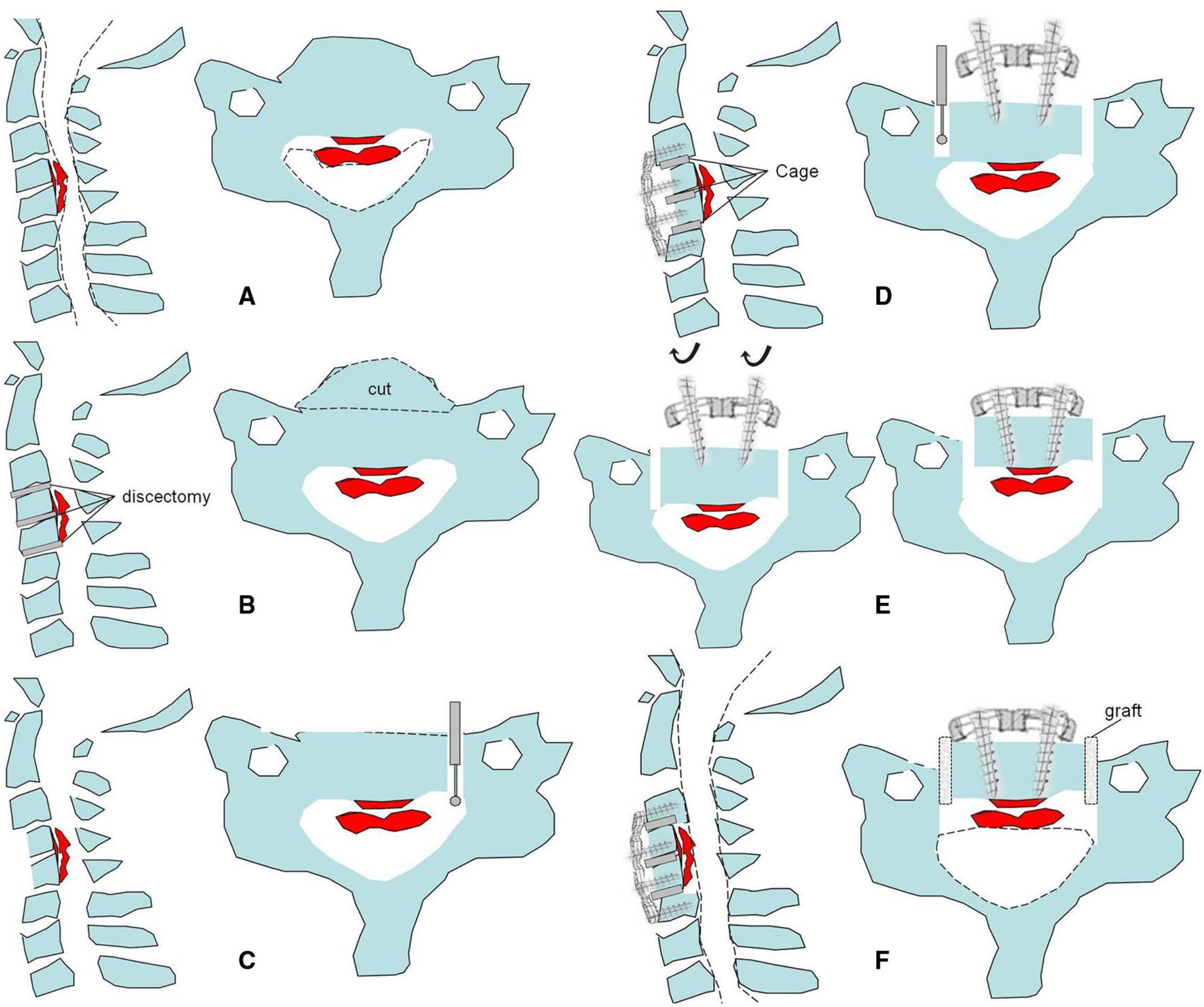


Fig. 2 ACAF surgical technique (diagram). Sagittal (left) and transverse (right) sectional images of the spine showing. **a** Two-level OPLL with DO from C4 to C5 (left) and double-layer sign on axial CT images (right). **b** Discectomy from C3–4 to C5–6 (left) and cutting of the anterior part of the vertebra (C4 and C5) according to the thickness of the ossified mass (right). **c** On the left side of the vertebra, creating a groove about 3 mm wide to the posterior wall of the vertebrae from C5 to C4. **d** Filling the intervertebral space from C5–6 to C3–4 with a cage filled with autologous bone fragments and placing an anterior plate on C3 and C6 as well as a screw in the central (C4 and C5) vertebrae (left) and then creating groove on the right side from C5 to C4 (right). **e** Axial views of before (left) and after (right) tightening of the screw in the middle vertebra (C4 and C5). **f** Sagittal (left) and transverse (right) views of the spinal canal showing the preserved ossified ligaments and DO moving forward with a good canal volume

Results

Perioperative characteristics

Perioperative details, including operative duration, blood loss, and hospital stay for each group are presented and compared between groups in Table 1. There was no significant difference in operative duration and blood loss between the two groups, but the hospital stay was significantly longer in the ACCF group ($P<0.05$).

Radiographic results

There was no significant difference in occupying rate, type and extent of OPLL, or type of double-layer sign between the two groups. On cross-sectional CT, decompression width and postoperative spinal canal area were both significantly larger in the ACAF group than in the ACCF group (18.0 ± 1.1 mm vs. 15.2 ± 0.9 mm, $P<0.01$, and 172.5 ± 31.7 mm^2 vs. 140.7 ± 26.9 mm^2, $P<0.01$, respectively). The AP diameter of the spinal cord on cross-sectional MRI was significantly greater in the ACAF group than in the ACCF group (5.5 ± 0.6 mm vs. 5.0 ± 1.1 mm, $P<0.05$).

Neurological outcomes

The mean postoperative JOA scores were significantly better than preoperative scores in the ACAF and ACCF groups ($P<0.01$, both; Table 1). However, the improvement rate was significantly better in the ACAF group than in the ACCF group ($82.4 \pm 8.8\%$ vs. $71.9 \pm 12.4\%$ $P<0.05$).

CSF leakage and treatment

Cerebrospinal fluid leakage was the main postoperative complication in this series. In the ACCF group, seven (22.6%) patients had dural tears, which were all diagnosed intraoperatively because of clear fluid extravasating from the wound. However, only one (3.6%) patient presented with CSF leakage in the ACAF group. This difference was significant ($P<0.01$).

If CSF leakage occurred, a drainage tube was pulled out in 12 h after operation, and continuous pressure was applied to the wound. Two patients received lumbar cisterna drainage. All patients were confined to bed rest with the head elevated 30° until CSF leakage ended. Of the seven patients with a cerebrospinal fluid leakage in the ACCF group, three (42.9%) had resolution of all signs and symptoms of the leak within 15 days; the other four (57.1%) within 30 days. For the patient with CSF leakage in the ACAF group, continuous pressure to the wound and suction via syringe were performed. CSF leakage disappeared in 10 days. No CSF leakage existed with infection.

Case report

Case 1

A 58-year-old man complained of numbness in both hands associated with slight gait disturbance for 6 months. Radiologic examination is shown in Fig. 3. ACAF of C4 (2 intervertebral disks) was performed. After the operation, the patient showed good neurological improvement from the myelopathy. His preoperative JOA score was 10 and postoperative JOA score at last follow-up was 16, for an improvement rate of 85.7%. There was no CSF leakage or other complications during the treatment.

Case 2

A 55-year-old woman complained of limb numbness and spastic weakness of the upper limb and hand for 2 years. The symptoms gradually increased in severity and coincided with gait disturbance during the year prior to presenting. Radiologic examination is shown in Fig. 4. ACAF of C3, C4 and C5 (four intervertebral disks) was performed. After the operation, the patient showed marked recovery from the myelopathy. Her preoperative JOA score was 7 and JOA score at last follow-up was 16, for an improvement rate of 90%. The 6-month-postoperative X-ray, CT, and MRI showed good decompression of the spinal cord and solid fusion. There was no CSF leakage or other complications during the treatment.

Discussion

OPLL, characterized by heterotopic bone formation in the posterior ligament, is a common cause of spinal stenosis with myelopathy [16]. Conservative treatment can lead to progression of related disabilities, and surgical intervention has been proven to be the most effective treatment [17–19]. Several surgical options for cervical OPLL have been established, which involve mainly anterior surgery (ACCF) or posterior surgery (laminoplasty and laminectomy with fusion). Although the optimum method for the treatment of cervical OPLL is controversial, anterior ACCF is recommended for treatment of massive OPLL or kyphotic alignment when the involved surgical segments are <3 levels [20–22]. In our previous study, we performed a prospective nonrandomized controlled clinical study with patients with multilevel cervical myelopathy due to OPLL and compared the therapeutic efficiency of laminoplasty and ACCF in the

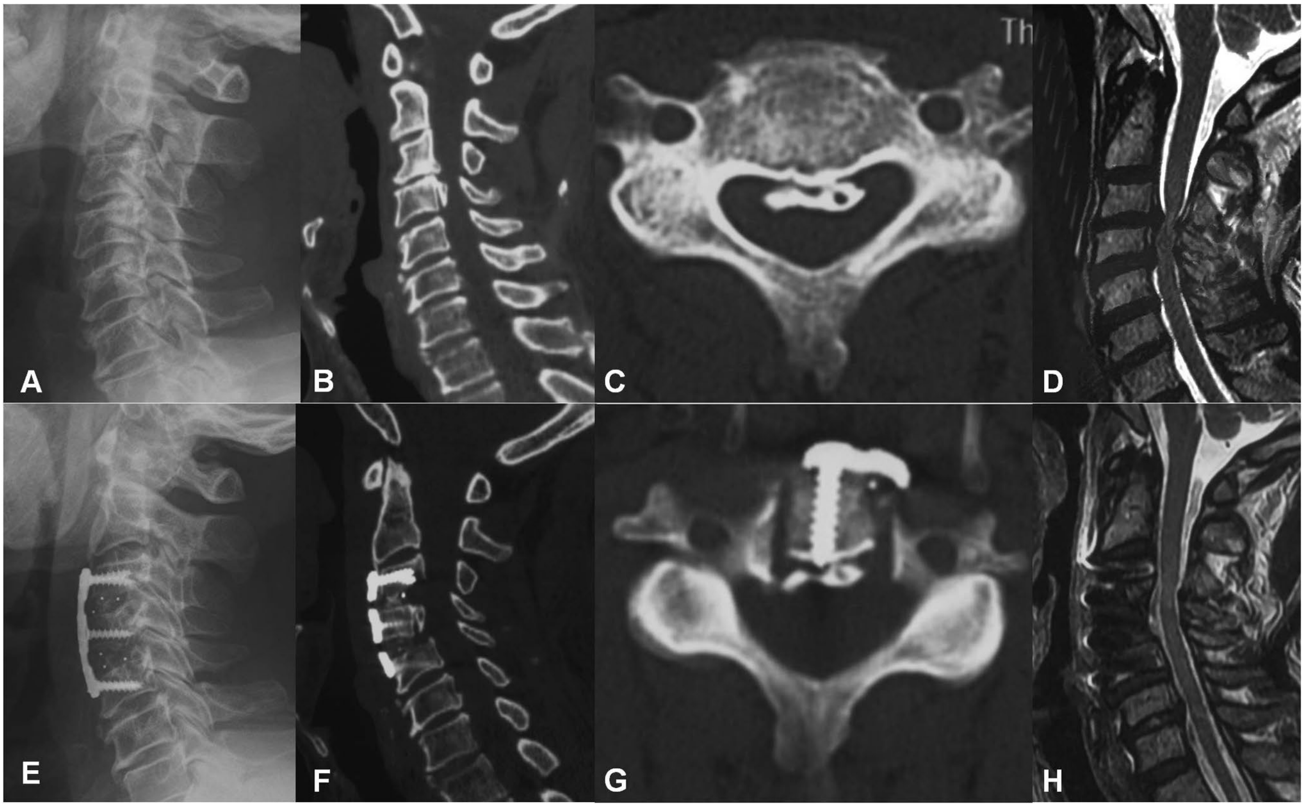


Fig. 3 A 58-year-old man undergoing ACAF. **a** Lateral X-ray and **b** sagittal CT of the cervical spine showing OPLL and DO at C4 level. **c** Axial CT showing double-layer sign of type C with the occupying rate of 35.7%. **d** Sagittal MRI showing severe spinal cord compression at C4 level. **e** Lateral X-ray and **f** sagittal CT showing ACAF of C4 with good spinal canal volume. **g** Axial CT showing the preserved ossified ligaments and DO moving forward with a well decompression width (18.1 mm) and spinal canal area (238.6 mm^2). **h** Sagittal MRI demonstrating good morphology of the spinal cord with no compression

management of multilevel cervical OPLL. We found that the anterior approach with relatively lower incidence of postoperative complications could be a better choice for cases with poor cervical curvature [23]. Wang also claimed that though the incidence of complications of anterior surgery is higher than that for posterior surgery, direct anterior decompression is advised when the complications can be controlled by advanced surgical techniques [24].

CSF leakage was one of the most common complications when ACCF was used to treat OPLL. DO and adhesion with OPLL was the main reason [1–4, 25]. In the present study, DO was evaluated via the double-layer sign, which is characterized by anterior and posterior ossified rims separated by hypertrophied posterior longitudinal ligament (PLL). It was more specific for DO because it was found in 10 of 12 patients. Mizuno et al. and Min et al. agreed that Japanese or Korean patients presenting with an ossified mass and double-layer sign on CT scans have an increased possibility of DO than those with single-layer signs (or en bloc type) [1, 26]. Chen reported that the specificity of double-layer sign appeared in more than 96% of patients with DO [2]. We also made a classification to double-layer sign and found that OPLL with double-layer sign of type C is almost inevitably followed by CSF leakage after anterior decompression [9]. Therefore, we used the double-layer sign as the diagnosis criteria of DO.

The aim of ACCF was to cut and remove the ossified ligaments, so DO and adhesion with OPLL must be confronted. Mizuno et al. described a surgical technique for DO associated with OPLL, in which the OPLL was separated from DO through a thin layer comprising a nonossified degenerated PLL, resulting in the avoidance of dural defect and CSF leakage [1]. We also proposed the “safe space” made by the specialized microdissector in the anterior cervical procedure for OPLL with DO [9]. However, a nonossified PLL layer and successful separation were not always present; in this case, a dural defect and uncontrolled CSF leakage usually occurred [1–3, 5, 8, 9]. Although CSF leakage normally did not affect neurological improvement, it led to a long hospital stay, prolonged recovery duration, high economic cost, higher infection probability, and increased chance of revision surgery [3, 9, 10]. In the reported studies, the recovery time of CSF leakage ranged from 1 week to 3 months [3, 7, 10]. Various secondary complications associated with a

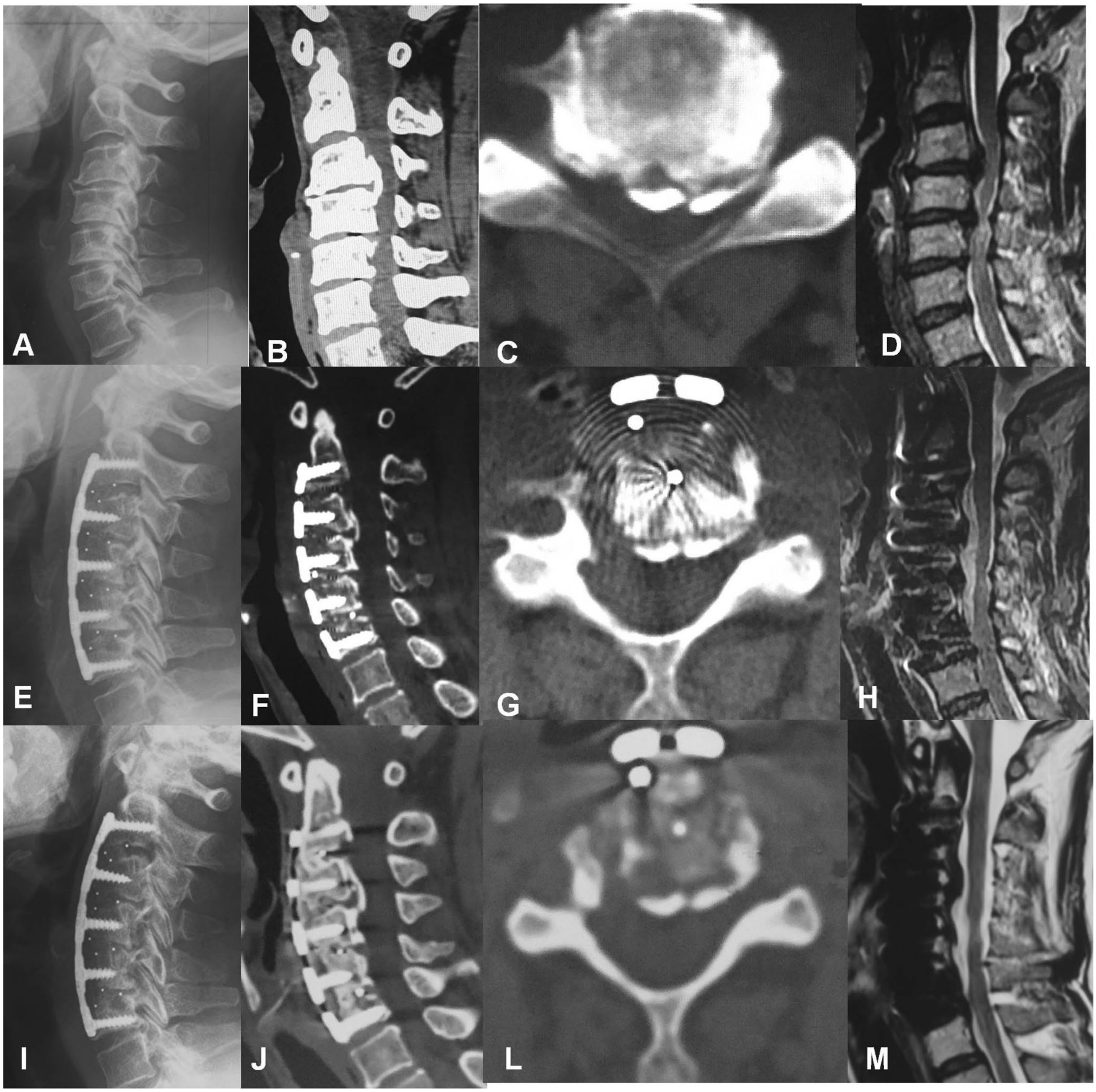


Fig. 4 A 55-year-old woman who underwent ACAF. **a** Lateral X-ray and **b** sagittal CT of the cervical spine showing spinal stenosis from C3 to C5 due to OPLL and DO, **c** Axial CT showing double-layer sign of type C with the occupying rate of 40.2%. **d** Sagittal T2-weighted MRI showing anterior spinal cord compression from C3 to C5 with increased signal intensity. **e** Lateral X-ray and **f** sagittal CT showing ACAF of C3, C4 and C5 with satisfied spinal canal volume. **g** Axial CT image showing the preserved ossified ligaments and DO moving forward with a well decompression width (20.2 mm) and spinal canal area (171.5 mm^2). **h** Sagittal MRI demonstrating good morphology of the spinal cord with no compression. **i** Lateral X-ray and **j** sagittal CT images at 6-month follow-up showing solid fusion. **l** Axial CT and Sagittal MRI at 6-month follow-up showing adequate decompression of the spinal cord

CSF leakage after cervical operations, including meningitis, delayed wound healing, airway obstruction, cutaneous CSF fistula, and pseudomeningocele, also appeared [11–13]. It is imperative to propose a novel technique to prevent CSF leakage. Our new surgical technique achieved adequate cord decompression and good neurological recovery not by cutting and removing the ossified ligaments directly, but by isolating the ossified mass and moving the vertebrae with ossified mass forward en bloc. This technique avoided disturbance to the DO and avoided CSF leakage. Only one patient underwent ACAF presented with CSF leakage in our study, which might be related to over-antedisplacement of the ossified mass. However, the incidence rate was much higher in the ACCF group, which also had a longer hospital stay. Second, the decompression width, area of the spinal canal, and AP diameter of the spinal cord were all lager in the ACAF group than in the ACCF group. An adequate decompression to the spinal cord enabled a better neurological recovery.

The present study had some limitations. The number of patients was limited and the follow-up period was relatively short. A study with more cases and longer follow-up duration should be performed. Additionally, this is a retrospective controlled study in a single center; a multicenter prospective randomized controlled study should be performed.

Conclusions

ACAF can reduce the occurrence of CSF leakage by moving the vertebrae with ossified ligaments and DO forward en bloc and simultaneously achieve adequate cord decompression and good neurological recovery. It provided an ideal option in the anterior treatment for OPLL with DO.

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Compliance with ethical standards

Conflict of interest None of the authors has any potential conflict of interest.

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