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One-stage combined decompression for cervical ossification of the posterior longitudinal ligament combined with upper thoracic ossification of the ligamentum flavum: Case report

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Abstract

Background and objective: Ossification of posterior longitudinal ligament (OPLL) and ossification of ligamentum flavum (OLF) are common clinical heterotopic ossification diseases and are the main causes of cervicothoracic spinal stenosis and spinal cord damage. However, the incidence of cervical OPLL combined with thoracic OLF is unclear, this clinical condition is difficult to treat, and the surgical approach is not fully established. We report two cases of patients with cervical OPLL combined with upper thoracic OLF treated with one-stage combined decompression.

Methods: The clinical data of 2 patients diagnosed with cervical OPLL combined with upper thoracic OLF (T1-3) and one-stage surgical treatment from January 2015 to January 2019 were analyzed.

Results: Two patients underwent one-stage combined decompression surgery, and the surgery was successfully completed, and there were no postoperative complications such as cerebrospinal fluid leakage and neurological deterioration. One patient's JOA score increased from 5 points before surgery to 11 points at 2 years of follow-up, and the improvement rate of JOA score was 50%; Another patient's JOA score increased from 11 points before surgery to 15 points at 2 years of follow-up, and the improvement rate of JOA score was 66.7%. Two patients were satisfied with postoperative recovery.

Conclusion: There are few reports on cervical OPLL combined with upper thoracic OLF. We report two cases of patients with cervical OPLL combined with upper thoracic OLF treated with one-stage combined decompression. The postoperative results were satisfactory. However, there were only 2 patients in this study and the follow-up period was short, in the future, high-quality studies with large samples, randomization, and long-term follow-up are still needed to further verify our conclusions.

Keywords: One-stage; Combined decompression; OPLL; TOLF; Spine surgery.

Abbreviations: OPLL: Ossification of the posterior longitudinal ligament; TOLF: Thoracic ossification of ligamentum flavum; JOA: Japanese Orthopaedic Association; CT:Computed Tomography; MRI: Magnetic Resonance Imaging.

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Introduction

Ossification of the spinal ligaments (OSL) is a pathological condition characterized by heterotopic ossification of ligaments around the spine, including ossification of posterior longitudinal ligament (OPLL), ossification of ligamentum flavum (OLF), ossification of anterior longitudinal ligament (OALL), ossification of nuchal ligament (ONL), and diffuse idiopathic skeletal hyperostosis, DISH), etc [1]. Among them, ossification of posterior longitudinal ligament (OPLL) and ossification of ligamentum flavum (OLF) are common clinical heterotopic ossification diseases, which are the main causes of cervicothoracic spinal stenosis and spinal cord damage [2-4]. However, the incidence of cervical OPLL combined with thoracic OLF is unclear [5]. This clinical condition is difficult to treat and the surgical approach is not fully established, we report two patients with cervical OPLL combined with upper thoracic OLF treated with one-stage combined decompression.

Materials and methods

Two patients were included in the study, and the average follow-up time was 33 months. The Japanese Orthopaedic Association (JOA) score was used to evaluate neurological function [6]. JOA improvement rateis considered to describe the function improvement after surgery, with the result of 100% considered to be all healed, 60–100% to be remarkable, 25–60% to be effective, less than 25% to be generally ineffective compared with preoperative values. This study was carried out with the approval of the ethics committee of our hospital, and written informed consent was obtained from all participants.

Operation method

Two patients underwent cervical posterior open-door laminoplasty (C3-7)+ thoracic posterior laminectomy, and the operations were performed by the same group of operators. Intraoperative application of motor-evoked potentials(MEPs) and somatosensory-evoked potentials (SEPs) monitoring.

The patients were placed in the prone position while under general anesthesia. Through bilateral paraspinal muscle dissection and the supraspinous ligaments entirely preserved, the interspinous ligaments were cut at the superior and inferior ends of the C3-7 levels. A full-thickness trough was drilled at the junction of the lateral mass and the lamina with a highspeed burr on the right side as the open-door, and a partial-thickness trough was drilled on the left side as the door-hinge. The lamina was elevated from the opendoor toward the hinge side for approximately 8 to 10 mm and stabilized with 8 or 10 mm miniplates and screws. After the cervical spine surgery is completed, continue to separate the skin and subcutaneous tissue layer by layer along the spinous process, exposing the spinous process, lamina and articular process and other structures. First, the spinous process and the outer layer of the lamina were bited off, and then the lateral lamina were removed by high-speed grinding drill in the longitudinal direction at the inner edge of the facet joints of the bilateral lamina. Then, the interspace of the lamina was enlarged, and the ligaments between the upper and lower spine were removed, and then used a high-speed grinding drill to scan the middle lamina with normal saline flushing, and did not polish each line too deeply until the ossified tissues were eggshell-like translucent. The remaining thin layer of bone was lifted with towelet forceps, and the inner vertebral plate, ossified ligamentum flavum and dural mater were gently probed with nerve exfoliator to see if there was adhesion. If there was no adhesion, the inner vertebral plate and ossified ligamentum flavum would be removed to achieve full decompression. Continued to nibble on both sides to the outer 1/3 of the facet joint, exposed both sides of the dural sac, at this time the dural sac could be completely expanded. After decompression was completed, rinse the operation field thoroughly, indwelling negative pressure drainage tube, suture the incision layer by layer, and the operation was finished. All patients were routinely treated with antibiotics within 3 days after operation. 7 days after operation, patients were encouraged to wear braces to move under the ground, and brace protection was maintained for about 3 months.

Results and illustrative cases

The two patients were followed up for more than 2 years, with an average follow-up time of 33 months.

Case 1

A 73-year-old male patient was admitted to the hospital mainly due to numbness and weakness of the limbs and a feeling of stepping on cotton in both lower limbs for half a year. The patient was previously healthy, and the above symptoms aggravated 2 months ago, and he developed urination disorder. Physical examination revealed sensory plane below the xiphoid process, bilateral Hoffmann's sign (+), bilateral knee tendon hyperreflexia, and bilateral Babinski sign (+).

Sagittal MRI showed C3-7 posterior longitudinal ligament ossification, T1-3 ligamentum flavum ossification, and spinal cord compression. Axial CT can clearly show that the ossified longitudinal ligament protrudes into the spinal canal with a high-density shadow on the posterior edge of the vertebral body, and the ossified ligamentum flavum shows a high-density shadow on one or both sides of the anterior edge of the lamina protruding into the spinal canal (Figure 1, Figure 2).



Figure 1: Preoperative MRI and CT of cervical spine Figure A shows sagittal MRI showing C3-7 posterior longitudinal ligament ossification and spinal cord compression, and Figures B, C, D, and E show axial CT can clearly show that the ossification of the posterior longitudinal ligament presents a high-density shadow at the posterior edge of the vertebral body and protrudes into the vertebra inside the tube.



Figure 2: Preoperative MRI and CT of thoracic spine

Figure A shows sagittal MRI showing T1-3 ligamentum flavum ossification and spinal cord compression, Figure B, C shows axial CT can clearly show that the ossified ligamentum flavum presents a high density shadow on one or both sides of the anterior edge of the lamina protruding into the spinal canal.

The patient was treated with one-stage operation of cervical posterior open-door laminoplasty (C3-7) + thoracic posterior laminectomy. Intraoperative application of motor-evoked potentials(MEPs) and somatosensory-evoked potentials (SEPs) monitoring. The operation was successfully completed, and there were no postoperative complications such as cerebrospinal fluid leakage (Figure 3). The patient's JOA score increased from 5 points before operation to 11 points at 2-year follow-up, the improvement rate of JOA score was 50%, and postoperative recovery was satisfactory. 10 days after the operation, the CT scan showed that the cervical spinal canal was enlarged, the ossified ligamentum flavum of the thoracic spine was completely removed, and the spinal cord was significantly relieved (Figure 4).



Figure 3: Intraoperative images Figure A shows the application of a high-speed drill to thin the lamina and ossified ligamentum flavum during the operation, and Figure B shows that the cervical spinal canal is completely enlarged, the decompression is sufficient, and the thoracic dural sac can be fully inflated.



Figure 4: Postoperative CT of cervical and thoracic spine Sagittal CT (Figure A) and axial CT showed that the cervical spinal canal was completely enlarged and decompressed sufficiently (Figures B, C, D, E), the ossified ligamentum flavum of the thoracic spine was completely removed, and the spinal cord compression was significantly relieved (Figures F, G); the patient's JOA score increased from 5 points before operation to 11 points at 2-year follow-up, the improvement rate of JOA score was 50%, and the postoperative recovery effect was satisfactory.

Case 2

A 65-year-old female patient was admitted to hospital mainly due to numbness of both upper extremities, weakness of both lower extremities, and unstable walking for 2 years. The patient had a 5-year history of hypertension. Physical examination revealed sensory plane below the both nipples, bilateral Hoffmann's sign (+), bilateral knee tendon hyperreflexia, and bilateral Babinski sign (+). Lateral X-ray of the cervical spine shows ossification shadow of posterior longitudinal ligament(Fig. 5). Sagittal MRI showed C3-7 posterior longitudinal ligament ossification, T1-3 ligamentum flavum ossification, and spinal cord compression. Axial CT can clearly show that the ossified longitudinal ligament protrudes into the spinal canal with a high-density shadow on the posterior edge of the vertebral body, and the ossified ligamentum flavum shows a high-density shadow on one or both sides of the anterior edge of the lamina protruding into the spinal canal (Figure 6).



Figure 5: Preoperative X-ray of cervical spine Lateral X-ray of the cervical spine shows ossification shadow of posterior longitudinal ligament.



Figure 6: Preoperative MRI and CT of cervical and thoracic spine Figure A shows sagittal MRI showing C3-7 posterior longitudinal ligament ossification, T1-3 ligamentum flavum ossification, and spinal cord compression; Figures B, C, D, and E show that the axial CT of the cervical spine can clearly show that the ossified posterior longitudinal ligament presents a high-density shadow at the posterior edge of the vertebral body and protrudes into the spinal canal; Figures F and G show that the axial CT of the thoracic spine can clearly show that the ossified ligamentum flavum presents a high-density shadow on one or both sides of the anterior edge of the lamina protruding into the spinal canal.

The patient was treated with one-stage operation of cervical posterior open-door laminoplasty (C3-7) + thoracic posterior laminectomy. Intraoperative application of motor-evoked potentials(MEPs) and somatosensory-evoked potentials (SEPs) monitoring. The operation was successfully completed, and there were no postoperative complications such as cerebrospinal fluid leakage (Figure 7). The patient's JOA score increased from 11 points before operation to 15 points at 2-year followup, the improvement rate of JOA score was 66.7%, and postoperative recovery was satisfactory. 10 days after the operation, the CT scan showed that the cervical spinal canal was enlarged, the ossified ligamentum flavum of the thoracic spine was completely removed, and the spinal cord was significantly relieved (Figure 8).



Figure 7: Intraoperative images

Figures A and B show the monitoring of motor evoked potentials (MEPs) and somatosensory evoked potentials (SEPs) during operation; Figure A shows intraoperative MEPs monitoring of the right lower extremity; Figure B shows intraoperative SEPs monitoring of the left upper extremity, and Figure C shows that the cervical spinal canal is completely enlarged, the decompression is sufficient, and the thoracic dural sac can be fully inflated.



Figure 8: Postoperative CT of cervical and thoracic spine Sagittal CT (Figure A) and axial CT showed that the cervical spinal canal was completely enlarged and decompressed sufficiently (Figures B, C, D, E), the ossified ligamentum flavum of the thoracic spine was completely removed, and the spinal cord compression was significantly relieved (Figures F, G); the patient's JOA score increased from 11 points before operation to 15 points at 2-year follow-up, the improvement rate of JOA score was 66.7%, and the postoperative recovery effect was satisfactory.

Discussion

Ossification of the spinal ligaments (OSL) is a pathological condition characterized by heterotopic ossification of ligaments around the spine, including ossification of posterior longitudinal ligament (OPLL), ossification of ligamentum flavum (OLF), ossification of anterior longitudinal ligament (OALL), ossification of nuchal ligament (ONL), and diffuse idiopathic skeletal hyperostosis, DISH), etc [1]. Among them, ossification of posterior longitudinal ligament (OPLL) and ossification of ligamentum flavum (OLF) are common clinical heterotopic ossification diseases, which are the main causes of cervicothoracic spinal stenosis and spinal cord damage [2-4]. However, the incidence of cervical OPLL combined with thoracic OLF is unclear [5]. It has been reported in the literature that the proportion of patients with the cervical OPLL combined with thoracic OLF ranges from 5.99% to 7.36% [7]. Park et al. [5] found that 33.8% of cervical OPLL patients had thoracic tandem ossification. A prospective study by Kawaguchi et al. [8] included 178 patients with symptomatic cervical OPLL, and 53.4% had thoracolumbar ligament ossification. Fujimori et al. [9] reported that in the general Japanese population, 13% of the cervical OPLL combined with thoracic OPLL, and 34% of the cervical OPLL combined with thoracic OLF. A recent epidemiological study with a large sample size of the Chinese population found that among patients with cervical OPLL, 21% had thoracic OPLL, and 44% had thoracic OLF [10].

There are no consistent conclusions about the etiology, pathogenesis, incidence, and risk factors of cervicalthoracic spine ligament ossification, which need to be further confirmed by more basic and clinical studies [5,11,12]. It is unclear why cervical OPLL combined with thoracic OLF occurs more often in the upper thoracic spine than at the common thoracolumbar junction. In some previous studies, mechanical stress has been suggested to play an important role in the progression of TOLF, and excessive activity increases the stress on the ligamentum flavum at the lesion site, resulting in ligament ossification in this segment [13-16]. Our study found that in patients with cervical OPLL, OPLL itself may lead to limited range of motion at

the cervicothoracic junction and upper thoracic spine. Excessive activity increases the stress on the ligamentum flavum in the upper thoracic spine, leading to ligament ossification in this segment, which explains why cervical OPLL combined with thoracic OLF occurs more often in the upper thoracic spine than at the common thoracolumbar junction.

For patients with confirmed cervicothoracic ossification, if there is no contraindication to surgery, surgery should be taken as soon as possible in principle [17-19]. At present, it is still controversial whether one-stage combined decompression should be performed for patients with cervical OPLL combined with upper thoracic OLF. One-stage combined operation can relieve spinal cord compression on all diseased segments at one time, shorten the treatment period and reduce medical costs, and avoid the risk of reoperation in the short term, but it expands the scope of surgical damage, increases the difficulty of surgery and postoperative recovery time, while increasing the incidence of complications such as intraoperative nerve injury, postoperative hematoma, cerebrospinal fluid leakage and deep infection [20-22]. Hu et al. [23] performed one-stage combined decompression for patients with continuous ossification of the cervical spine and upper thoracic spine, and found that 37.5% of the patients developed serious complications such as cerebrospinal fluid leakage and neurological deterioration. We report two patients with cervical OPLL combined with upper thoracic OLF who underwent one-stage combined decompression. The operations in both cases were successfully completed, and there were no postoperative complications such as cerebrospinal fluid leakage and neurological deterioration. One patient's JOA score increased from 5 points before surgery to 11 points at 2 years of follow-up, and the improvement rate of JOA score was 50%; Another patient's JOA score increased from 11 points before surgery to 15 points at 2 years of follow-up, and the improvement rate of JOA score was 66.7%. Two patients were satisfied with postoperative recovery. One-stage combined surgery or staged surgery for patients with cervical OPLL combined with upper thoracic OLF depends on many factors, including the severity of the lesion and the patient's ability to accept it, etc. Adequate preoperative evaluation is crucial for the selection of surgical methods.

Conclusion

There are few reports on cervical OPLL combined with upper thoracic OLF. We report two cases of patients with cervical OPLL combined with upper thoracic OLF treated with one-stage combined decompression. The postoperative results were satisfactory. However, there were only 2 patients in this study and the follow-up period was short, in the future, high-quality studies with large samples, randomization, and long-term follow-up are still needed to further verify our conclusions.

Declarations

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Ethics approval and consent to participate: This article does not contain any studies with human participants or animals performed by any of the authors.

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