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Case report

Anterior spinal cord herniation after multilevel anterior cervical corpectomy: A case report

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ABSTRACT

Many complications related to the resection of an ossified posterior longitudinal ligament via the anterior approach have been reported. Postoperative neurological deterioration is one such complication that may appear due to massive anterior spinal cord herniation related to a dural defect following resection of the ossified posterior longitudinal ligament. Specifically, spinal cord herniations have been reported to be associated with posterior approaches, and a large number of theories regarding this association have been offered by various authors. However, anterior spinal cord herniation is extremely rare, and its pathophysiology has not yet been explained. In this case report, we report a male patient who experienced anterior spinal cord herniation following anterior surgery. Spinal cord herniation may develop following the removal of the anterior cervical corpectomy. Therefore, surgeons should be aware of this condition when planning treatments for cervical spondylotic myelopathy via the anterior approach.

1. Introduction

Various complications associated with the treatment of cervical spondylotic myelopathy due to ossified posterior longitudinal ligament (OPLLs) have been reported [1–3]. The most common complications are inadequate decompression, haemorrhage, postoperative neurological deterioration and cerebrospinal fluid (CSF) leakage [3–5].

Spinal cord herniation (SCH) is frequently idiopathic or spontaneous. Additionally, trauma and surgery may be factors that are related to the development of SCH [6,7]. However, SCH following cervical anterior multilevel corpectomy for the treatment of cervical spondylotic myelopathy due to OPLL has rarely been reported [4], and the cause of this pathophysiology have not yet been explained. Therefore, we present a case who developed anterior SCH following two-level corpectomy and OPLL resection and explain the pathophysiology of

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Abbreviations: OPLL, ossified posterior longitudinal ligament; CSF, cerebrospinal fluid; SCH, spinal cord herniation; MR, Imagnetic resonance imaging; COPD, chronic obstructive pulmonary disease.

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the formation of the SCH via a new hypothesis in the present report.

2. Case report

A 60-year-old male patient presented with a three-year history of quadriparesis and neck pain. A neurological examination revealed that his muscle strengths were 4/5 in the bilateral upper extremities and 3/5 in the lower extremities. Deep sensory loss was present in the bilateral upper and lower extremities, and the deep tendon reflexes were hyperactive in the lower extremities but hypoactive in the upper extremities. Pathological reflexes were identified in the bilateral lower extremities. Magnetic resonance imaging (MRI) and computed tomography revealed an OPLL that caused cord compression between the C3 and C6 disc levels. The lesion was in the midline of the vertebral body (Fig. 1). The most severe compression occurred at the C5–6 levels (Fig. 2A and B). We decided on anterior spinal surgery, but the patient had chronic obstructive pulmonary disease (COPD) and was therefore referred to a chest disease doctor prior to surgery. Medical treatment was administered for two weeks prior to the surgery. Approval forms were collected from the patient before the surgery. During the operation, C4 and C5 vertebral body resections were performed with Kerrison punch and a high-speed drill. Subsequently, the central portion of the OPLL was removed with a diamond drill, but a tiny shell of the cortical bone was left in place. While we were removing this central portion of the OPLL using a micro-hook and Kerrison punch, the dura was torn, and the arachnoid membrane and neural tissue were directly exposed. Direct repair of the dura was not possible. Therefore, the dura was closed using a synthetic dural graft and tissue adhesive, and an iliac bone autograft was subsequently inserted into the corpectomy side. A cervical plaque was inserted and screwed. The patient's neurological status was examined on the first postoperative day, and no neck pain was noted, and his upper and lower extremities were relaxed. However, he had a persistent cough during the postoperative period. CSF collection was performed under the incision field on the second postoperative day. To achieve this collection, an external lumbar drainage catheter was placed at the lower lumbar level. The lumbar catheter was withdrawn on the fourth postoperative day because no CSF was collected under the incision site. The patient's neurological examination results worsened on the sixth postoperative day. The

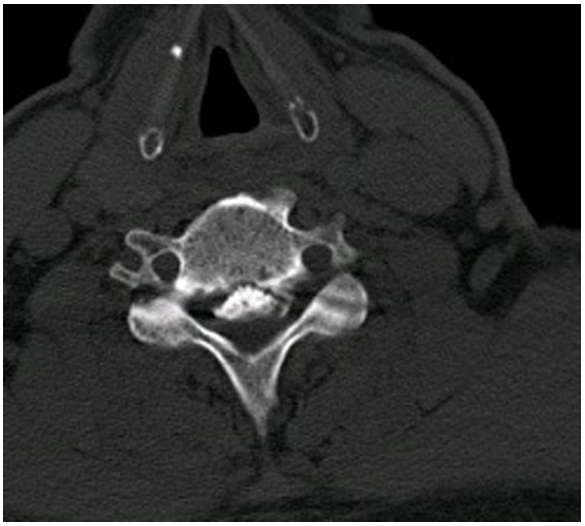

Fig. 1 – Preoperative cervical axial CT showing the location of the OPLL mass on the midline of the cervical spine.

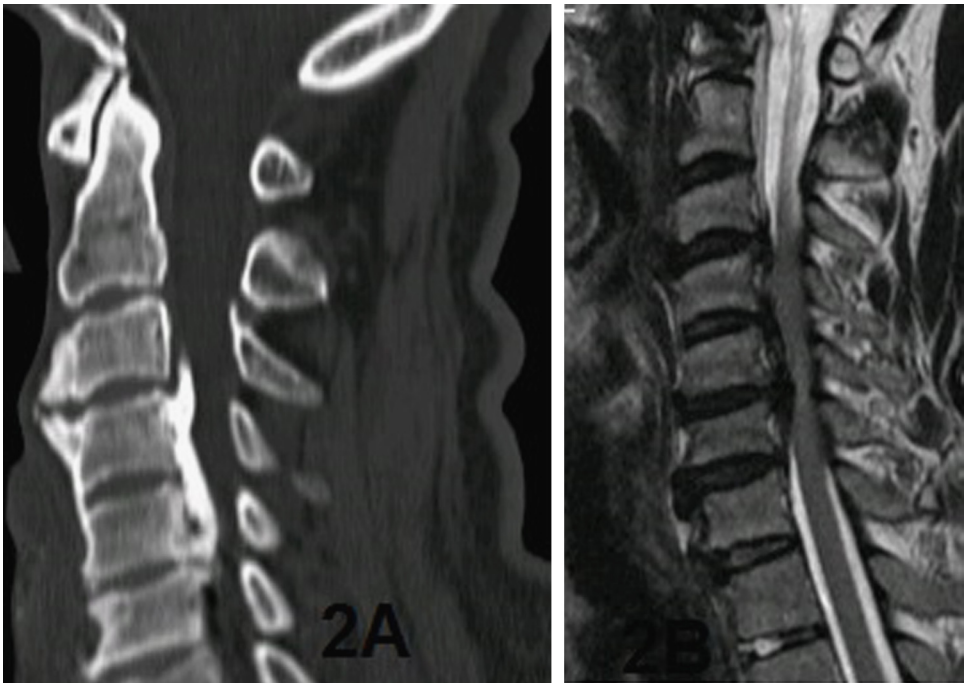

Fig. 2 – The maximal compression area occurred at the C5–C6 levels of the cervical spine as indicated by preoperative sagittal CT (A) and sagittal T2-weighted MRI images (B).

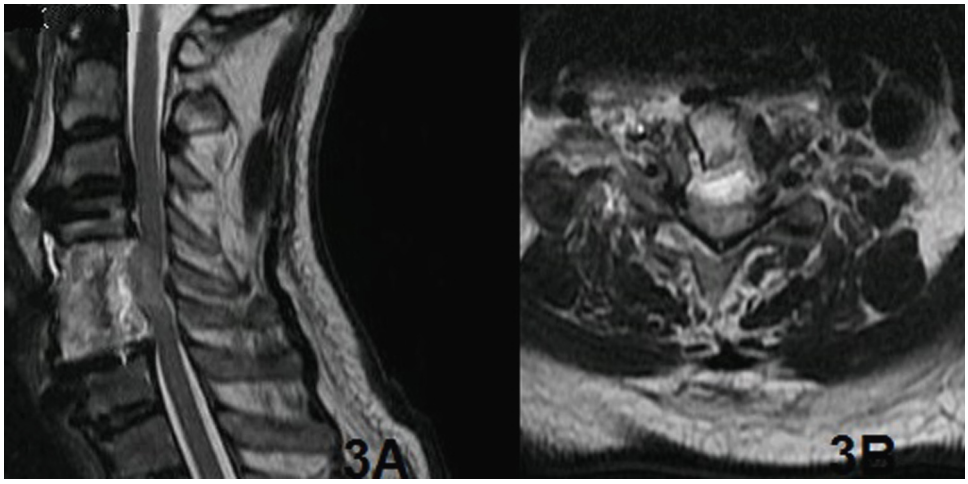

Fig. 3 – In the first postoperative sagittal T2-weighted MRI image following the anterior resection of the OPLL, an anterior spinal cord herniation was detected (A). The first axial T2-weighted MRI image showing the insertion of an iliac bone graft and adequate decompression of the ventral surface of the cervical spinal cord via the anterior cervical corpectomy (B).

patient underwent an urgent MRI that revealed that the cord had herniated into the corpectomy site and that edema into the spinal cord was present (Fig. 3A and B). We began high-dose methyl prednisolone infusion. We performed an additional MRI, which revealed that the spinal cord had remained herniated into the corpectomy side, the posterior subarachnoid space had expanded, and the edema in the spinal cord had decreased (Fig. 4). The patient's neurological condition improved slightly during the follow-up.

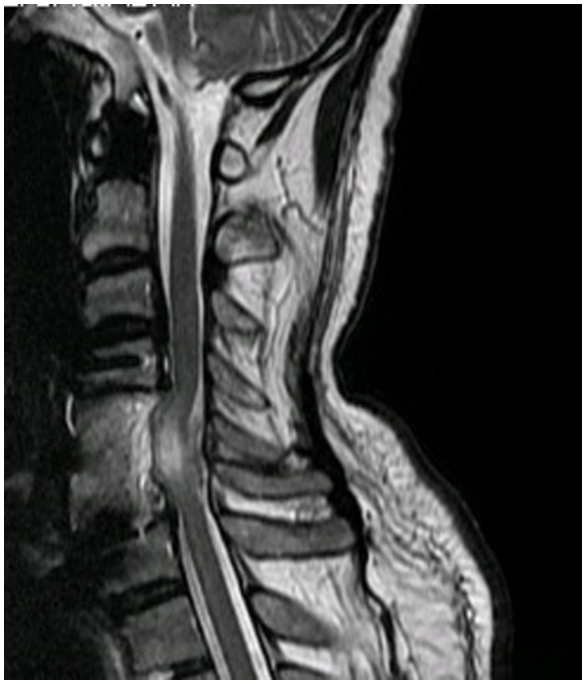

Fig. 4 – The second control sagittal T2-weighted MRI image showing that the posterior subarachnoid space was wide, and the edema of the spinal cord had decreased.

3. Discussion

SCHs are frequently idiopathic or occur spontaneously. However, SCHs may also result from surgery or trauma [6,7]. Therefore, SCHs can be classified as spontaneous or post-traumatic depending on etiology [6,8–10]. Idiopathic SCHs may frequently develop in the thoracic region, on the ventral surface, and occasionally on the ventrolateral surface [7,11–13]. Iatrogenic SCHs are associated with the posterior approach, and all reported cases of such herniations have been located in the dorsal region and frequently occur together with the formation of a pseudomeningocele [6,8,12,13].

Many theories have been proposed to explain the pathophysiology of SCHs. Kumar et al. reported that two factors coexist to enable the development of a SCH. The first is a dural defect that leads to a congenital arachnoid cyst or a pseudomeningocele, and the second is a defect that is located on the concave surface of the spinal curvature (i.e., dorsally in the cervical region and ventrally or ventrolaterally in the thoracic region). These authors reported that SCHs are rare because the likelihood of the coexistence of these two factors is low [12]. Martin explained that spinal cord herniations occur on the ventral surface in thoracic region because the thoracic curvature pulls the spinal cord in the anterior direction [14]. Another theory was proposed by Isu et al. In this theory, the authors stated that herniations could develop due to the cord being pushed from the ventral region into an enlarged arachnoid cyst in the posterior region of the cord [15]. Another theory is that cord herniations are congenital [12]. However, SCHs have not been incidentally reported in autopsies [12]. According to Nakazawa et al., SCHs might arise from duplications in the dura [16]. Another theory that was proposed by Najjar et al. is that an inflammatory process or an event might initially cause the spinal cord to adhere to the dura ventrally. Together with CSF pulsation, such an event may cause the erosion of the dura and the formation of a dural defect that would result in a SCH [13].

Many complications related with OPLL resection via anterior surgeries have been reported. Among these complications CSF leakage is the most common. The causes of dural defects during OPLL resection include the embedding of the mass of the OPLL in the dura, thinning of the dura, replacement of the dura, and the ossified mass. Therefore, CSF leakage develops due to dural and arachnoidal injuries during OPLL resection. Although many complications related to CSF fistulas have been reported, the complications related to CSF leakage, such as anterior spinal cord herniation, have not yet been reported. Therefore, there is no information about spinal cord herniation related to CSF leakage in the literature. However, SCH into the corpectomy field following OPLL resection has rarely been reported in the literature [4,17].

Anterior cervical spinal multilevel corpectomy and anterior cervical SCH following OPLL resection have been reported by Min et al. These authors reported that herniations of the cervical spinal cord into the corpectomy field result from cord herniation, the presence of a non-decompressed OPLL mass in the adjacent level, the presence of a diffuse dural defect (double layer sign) and the performance of multilevel anterior corpectomy. Additionally, these authors reported that anterior cervical SCHs develop due to causes opposite to those reported by Kumar [4].

We believe that the initiating event may be dural injury during the resection of an OPLL mass (double layer sign area) via anterior surgery. In patients with have respiratory diseases, such as COPD, symptoms that increase intraspinal pressure, such as coughing following extubation in the postoperative period, may facilitate the implantation of the arachnoid membrane into the dural defect area. Following the implantation of the arachnoid membrane into the dural defect area, some portion of the arachnoid may herniate into the corpectomy area from a dural defect. This system would then function as a one-way valve in which the CSF may enter but cannot leave. Spinal cord traction-related ischemia may lead to increased ischemia, and the softened cord may easily herniate into the low-pressure site in the anterior region. This process may resemble leptomeningeal cyst formation following calvarial diastatic fracture. Our opinion is that a non-decompressed adjacent level OPLL mass may be a causative factor in the development of spinal cord ischemia because spinal cord traction related to an anterior SCH may cause the compression of the anterior vascular structures of the spinal cord due to the mass in the adjacent level. Therefore, anterior spinal cord traction and adjacent non-decompressed OPLL mass compression together may be the factors that increase the occurrence of spinal cord ischemia.

The observation of a postoperative anterior SCH partially supports the literature because dural defects are possible in many theories. In the present case, the SCH was not on the concave surface and did not coexist with a pseudomeningocele. The frequencies of anterior and posterior surgeries may also affect the formation of SCHs on the ventral or concave surfaces. Additionally, a review of the literature is terms of the treatment of SCHs indicated that the repair of dural defects seems sufficient [2]. The present study and a study by Min et al. [4] revealed the same features, as the latter authors observed a OPLL mass and performed a two-level corpectomy via an anterior surgery that resulted in a dural injury, CSF leakage and the development of an anterior SCH. However, Min et al. elected to utilize a posterior decompression surgery for the treatment of the anterior SCH because a non-decompressed OPLL mass could potentially induce kinking site in the cord. However, this surgical approach is not supported by the literature. Posterior decompression surgical approaches, such as laminoplasty and laminectomy, might have been options for the treatment of the cervical spondylotic myelopathy following the anterior surgery in our case. However, we preferred the anterior surgical approach. Because the majority of the compressed area was on the ventral surface of the spinal cord and the posterior subarachnoid space was open in the adjacent level, there was a non-decompressed OPLL mass area due to the anterior SCH. Additionally, in our opinion, the initiating event may have been a dural injury that occurred during the resection of the OPLL mass. Therefore, if a lumbar drainage had been inserted in the early postoperative period, the anterior herniation of the spinal cord would not have developed because the intraspinal pressure would have been balanced. We also believe that the maintaining he lumbar drainage for as long as 5–7 days would have balanced the pressure and might have been clinically beneficial because the clinical symptoms of our patient began one day after the removal of the lumbar drainage. Therefore, the appropriate surgical treatment of anterior dural defect-related anterior SCHs is still open to debate.

In conclusion, anterior SCHs may develop following the treatment of OPLLs via anterior cervical corpectomy. Despite the rarity of this condition, it may lead to subacute postoperative neurologic deterioration. However, the causative factors and pathophysiology of anterior SCHs following anterior cervical corpectomy have not yet been clearly explained, and dural injury, COPD, non-decompressed adjacent level OPPL and anterior cervical multilevel corpectomy may be causative factors in the development of SCHs. Postoperative lumbar drainage application may be beneficial if it is inserted in the early postoperative period. Another option is the application of the floating corpectomy technique.

Conflict of interest

The authors declare that there is no conflict of interest.

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Regarding the making of this study, Zahir Kizilay was instrumental in endeavouring various works starting from designing the study, data collection with the help of Ali Yilmaz, literature search works with the help of Erdal Coskun, and evaluating and accepting the final manuscript with due help from Ozgur Ismailoglu and Erdal Coskun. Earlier Ali Yilmaz corroborated data interpretation works. We have no financial interest. This case report has not been published or presented in a meeting.

Ethics

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; Uniform Requirements for manuscripts submitted to Biomedical journals.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at doi:10.1016/j.pjnns.2015.11.009.

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