A Case Report of Possible Thoracic Interdural Ganglion Cyst

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Abstract

There are some intraspinal cystic lesions presenting with myelopathy. We report a case of myelopathy caused by a possible thoracic interdural ganglion cyst. A 70-year-old man with subacute bilateral lower extremity numbness, muscle weakness, and gait disturbance presented to our hospital. Magnetic resonance (MR) images showed a cystic lesion which compresses the left dorsolateral intraspinal space of T2-3. During surgery, a ganglion cyst was found without adhering to the periphery of the epidural space. The capsule and contents were removed. He showed postoperative improvement in activities of daily living. A postoperative pathological diagnosis of ganglion cyst was made. The development mechanism of thoracic interdural ganglion cysts is unknown. To our knowledge, this is the first report of this disease. Surgery improved symptoms of a patient with myelopathy caused by thoracic interdural ganglion cysts. This must be considered as one of the cystic lesions presenting with myelopathy.

Keywords: interdural cyst, ganglion cyst

Introduction

Sometimes, cystic lesions develop in the spinal canal, which mostly originates with the ligamentum flavum, posterior longitudinal ligament (PLL), and joint capsule. Here, we report our experience with the good surgical outcome of a rare case of myelopathy resulting from a possible thoracic interdural ganglion cyst. To the best of our knowledge, no study has been reported on thoracic interdural ganglion cysts.

Case Report

A 70-year-old man with subacute bilateral lower extremity numbness and gait disturbance presented to our hospital. Diminished superficial and deep sensations in the lower extremities, particularly in the left lower extremity, were observed, along with reduced tendon reflexes in the lower extremities. However, bladder and rectal disturbances were not identified. Manual muscle testing (iliopectineus, 3/3; quadriceps femoris, 4/4; tibialis anterior, 3/3; extensor hallucis longus, 3/2+; and gastrocnemius, 4/3) demonstrated muscle weakness, and modified Rankin Scale (mRS, a score of approximately 3) revealed gait disturbance. There was no appreciable disease in the family history. T2-weighted magnetic resonance (MR) images were taken in two planes (Fig. 1). The sagittal images (A and B) revealed displacement of a cystic lesion into the spinal canal at the left posterior epidural space of the second and third thoracic vertebrae (T2-3). On the axial image (C), inside of the cyst displayed high intensity signal, whereas the cyst wall had low intensity signal. Computed tomography (CT) scans revealed a low-density area inside the cyst, and there was neither a clearly calcified cyst nor a connection to bones located peripheral to the cystic lesion. Although epidural hematoma, hemorrhage of ligamentum flavum cyst, and hemorrhage into facet cyst were suspected, interdural cyst was not considered in the differential diagnosis.
The patient presenting with the symptoms of paralysis was admitted to our hospital and underwent surgery and received treatment. Under general anesthesia, a posterior spine midline incision was made in the prone position, followed by bilateral partial laminectomy at T2-3. A cystic lesion was identifiable after removal of the ligamentum flavum. However, the cyst was connected with neither the ligamentum flavum and joint capsule on the periphery nor the anterior components of thoracic vertebrae. The dura mater itself appeared rather swollen (Fig. 2). When a small incision was made in the lesion, xanthochromic viscous fluid flowed out of the cyst. The incised cyst wall appeared to be the dura mater, and the dura mater apparently extended over the incised cystic basement, instead of the arachnoid mater (Fig. 3). Fibrous tissue which proliferated around the cystic margin was immediately removed. If a definitive pathological diagnosis of suspicious for tumorous lesion was made after surgery, we decided to perform another surgery for removing the dura mater and repairing the area using artificial dura substitutes. Therefore, the dura-like membrane found in the cystic basement remained intact, and adipose tissue was grafted, followed by surgical incision closure.

After surgery, symptoms were gradually improved, and the patient was able to independently walk and improved from mRS 3 to 1. Pathological examination revealed that the cyst wall was composed of fibrous tissue in the absence of synovial layer. In the cyst wall, hyaline and mucous degeneration of collagen fibers was observed. Therefore, the cyst wall was considered to be consistent with a part of dura mater. In mucus within the cyst, hyaline and mucous degeneration of collagen fibers, along with chondroid metaplasia-like change, were observed. We presumed that a cystic lesion was caused by degeneration of collagen fibers, and the diagnosis of interdural ganglion cyst was made based on the region where the lesion existed (Fig. 4). No suspicious tumorous lesion was found; therefore, additional surgery was not performed to remove the dura mater. Postoperative MR images confirmed total removal of the lesion, and 2 years after surgery,
Among recurrent formation of interdural ganglion cyst has not been observed.

**Discussion**

A cystic lesion which is found in the intraspinal epidural space and develops adjacent to the intervertebral joint was called a synovial cyst or a ganglion cyst. In 1974, Kao et al. advocated the concept of juxta-facet cyst because no difference was observed in clinical prognosis between synovial and ganglion cysts. The formation of juxta-facet cysts, largely linked to degeneration, is mostly found in the lumbar vertebrae, particularly in high lumbar mobility at the fourth and fifth lumbar vertebrae and the fifth lumbar through first sacral vertebrae. Although both synovial and ganglion cysts have similar clinical symptoms (e.g., sciatica, intermittent claudication, and low back pain), differences exist between the two of them. While a synovial cyst is connected to the joint capsule and has a synovial lining, a ganglion cyst is not connected to the joint capsule and has the fibrous cyst wall without a synovial lining. Furthermore, a synovial cyst contains xanthochromic serous fluid, but a ganglion cyst contains viscous fluid.

Intraspinal cystic lesions include a ligamentum flavum cyst and a PLL cyst, and these cysts seem to arise from mucous degeneration of the ligament. These cysts, without an epithelial lining, protrude into the spinal canal, potentially causing nerve compression.

A ligamentum flavum cyst exists within the ligamentum flavum and is isolated without continuity with periphery. For these reasons, complete removal of the lesion is the best option to prevent a recurrence; however, if strongly adhering to the dura mater occurs, we decompress cyst contents and partially remove the layers within the cyst to prevent dural laceration and cerebrospinal fluid leak.

PLL cysts, which contain a jelly-like material, are found outside the dura mater located within the ventrolateral portion of the spinal canal on the posterior surface of the vertebral body. PLL cysts seem to develop due to repetitive trauma, joint disease, mucous degeneration, and spondylolisthesis, but they are distinguished from discal cysts which communicate with the intervertebral disc. Total surgical removal is the basic strategy for the treatment of PLL cysts, but CT-guided or endoscopic...
ultrasound-guided fine needle aspiration is also effective.\(^9\)

Although causes are unclear, discal cysts seem to develop due to a traumatic tear in the annulus fibrosus and the subsequently accumulated outflows, leading to reactive pseudomembrane formation and cyst development. On MR images, a discal cyst presents as a lobular cystic lesion attached to the epidural space and links to the intervertebral disc space via a stalk through an annular fissure.\(^10\) While a discal cyst shows low intensity signal on T1-weighted images, it shows high intensity signal on T2-weighted images. Discography demonstrates that a contrast agent injected into the intervertebral disc space leaks into the cyst, which elicits pain radiating to the lower extremity. Spontaneous regression of a discal cyst has been reported, but the use of epidural steroid and local analgesics is effective as symptomatic therapy. However, surgical removal is typically performed in the case of a severe symptom.\(^11,12\)

In our case, preoperative T2-weighted MR images showed a cystic lesion displaying high intensity signal at the left dorsal region of T2-3, and similarly, intraoperative finding showed that the lesion was located in the dorsal region. These findings demonstrated that there was no clear macroscopic connection between the cystic lesion and ventral structures. Furthermore, neither continuity with the ligamentum flavum nor discriminative defective regions in the dura mater were detected. Although the origin of intradural ganglion cysts is unknown, there seems to be a low possibility that an intradural ganglion cyst was caused by mucous degeneration of the dura mater. According to previous studies, there was a report on the formation of lumber intradural ganglion cyst presumably resulting from PLL cyst which invaded the dura mater.\(^13\) We also investigated the possibility of interdural invasion of other types of cystic lesions.

Based on our pathological findings, the cyst capsule was consistent with a part of dura mater. The mucus within the cyst along with chondroid metaplasia-like change was consistent with the findings of ganglion cyst. Regarding as the location of the cyst, because the outer cyst wall submitted to pathological examination was consistent of a part of dura mater, the cyst was located in the subdural space. The residual cystic basement was macroscopically also dura-like membrane, not arachnoid membrane. Therefore, the cyst was considered to be located in the interdural space, but this idea was a matter of speculation because the residual cystic basement was not examined pathologically. A case of arachnoid cyst between outer and inner dura was reported,\(^14\) suggesting the possibility of interdural invasion of other types of cystic lesions in our present case. From this point of view, it is difficult for identifying the cyst origin.

The cyst was considered to be discal cyst because of chondroid metaplasia-like change, but it was located in the dorsal region and isolated without continuity with the intervertebral disc and the periphery in our intraoperative findings. Facet cyst was considered to derive from cartilage component of facet joint, and it was possible that ganglion cyst occurred from synovial epithelium loss in the process of degeneration and destruction of synovial cyst,\(^15,16\) but the cyst also had no continuity with joint capsule. It was reported that chondroid metaplasia-like change of ligamentum flavum occurred by mechanical stress could happen in the process of degeneration\(^17\) so that ligamentum flavum was considered to be the cyst origin, but ligamentum flavum was macroscopically intact and had no adherence to the periphery.

The cyst in our present case had no continuity with epidural periphery, and healing process of the dura mater (the invaded region) without adhesion and scar was also unclear so that we could not conclude the cyst origin.

If continuity between the cyst and its origin was broken in the healing process of dural scar, it was most likely that ligamentum flavum cyst invaded into interdural space. We considered the possibility that a part of deep layer of ligamentum flavum in the cervicothoracic junction was degenerated by mechanical stress, and the cyst derived from ligamentum flavum invaded into interdural space. However, this idea was a matter of speculation, and further investigation was needed.

Regarding our surgical procedure, first, the duralike membrane found in the cystic basement remained intact. If a definitive pathological diagnosis of suspicious for tumor lesion was postoperatively made, we decided to perform another surgery to remove the dura mater and repair the area using artificial dura substitutes. Performing total removal of the dura mater (the lesion region) and duraplasty seems to be the right decision if a challenging intraoperative decision-making is anticipated due to a rare lesion found during surgery. To our knowledge, this is the first report of a thoracic interdural ganglion cyst.

**Conclusion**

We report a case of an elderly man with myelopathy caused by a thoracic interdural ganglion cyst of T2-3. Symptoms were improved after removal of a cystic lesion, and recurrence has not been observed.
The origin and cause of interdural ganglion cysts are still unknown, but when symptoms become worse, surgical removal seems to be optimal. This is extremely rare, but spine surgeons need to consider this case as a differential diagnosis of intraspinal cystic lesion at the thoracic level.

Conflicts of Interest Disclosure

The authors declare no conflicts of interest associated with this manuscript. The authors have no conflicts of interest directly relevant to the content of this article.

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