Atlantoaxial Intradural Synovial Cyst Mimicking an Extradural Lesion Adjacent to a Retro-odontoid Pseudotumor: A Case Report

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Abstract

Atlantoaxial synovial cysts can very rarely penetrate the dura mater into the thecal sac and cause direct neural compression. Several case reports have been available on “intradural synovial cysts” (IDSCs). In this study, we report on a case with an atlantoaxial IDSC mimicking an extradural lesion. A 90-year-old man was diagnosed with a cystic lesion located laterally to the atlantoaxial joint adjacent to the retro-odontoid pseudotumor (ROP) causing cervical spinal cord compression. Thus, surgical removal was planned. On preoperative examination, the cyst, which had a two-layer structure showing a T2-isointense small mass inside a T2-hyperintense lesion, was thought to be located in the extradural region. However, operative findings showed that the cyst was located inside the dura mater. Histopathological examination suggested a synovial cyst. No recurrence of the cyst was observed until the latest follow-up after 3 years, and the ROP decreased in size. Almost all IDSCs reported previously were observed in the medial site of the atlantoaxial joint. In our case, however, the cyst was observed adjacent to the posteromedial site of the right atlantoaxial joint and the ROP, mimicking an extradural lesion. We had no knowledge regarding the IDSC before the surgery and assumed an extradural lesion. Albeit rare, the existence of such a condition should be considered.

Keywords: Synovial cyst, Retro-odontoid pseudotumor, Intradural

Introduction

Synovial cysts are commonly observed as an extradural mass frequently arising from the lower lumber spine and occasionally from the cervical spine. It originates from the capsule and tendon sheath of the synovial joint. These cysts can occur at the craniovertebral junction, even without complicating diseases such as rheumatoid arthritis, which causes spinal instability.⁴,⁵,⁶ Approximately 70 cases with synovial cysts or ganglion cysts at the C1-C2 level have been reported previously, all of which were associated with trauma or atlantoaxial instability.⁵⁶ Evidence suggests that the cyst originates from the transverse ligament behind the dentate process of the axis.⁶⁷

Albeit quite rare, synovial cysts can penetrate the dura mater into the thecal sac and cause direct neural compression. Several case reports have been published on “intradural synovial cysts” (IDSCs).⁸,⁹,¹⁰ A review of these reports showed that dural penetration of the extradural cyst most frequently occurs at the craniovertebral junction. Neurological symptoms had been caused by direct compression of the spinal cord, brain stem, or hypoglossal nerve. This paper reports our experience with a case that developed IDSC at the atlantoaxial joints. The cyst mimicked an extradural cyst in preoperative imaging studies, and its intradural location had been noticed only during surgery. This paper aims to highlight the occurrence of these cases to guide future diagnoses of
IDSC. Written informed consent for publication of this report and accompanying images was obtained from the patient.

Case Report

A 90-year-old man presented with numbness in the right hand and motor weakness in the right arm and the right limbs. The progression of these symptoms prompted hospital visitation. The patient had undergone C3-C7 laminoplasty for cervical spondylotic myelopathy 4 years before. Flexion-extension cervical plain radiography showed no spinal instability but mild kyphosis with a decrease in the range of motion at the C3-C7. Magnetic resonance imaging (MRI) revealed a cystic mass occupying the right one-third part of the spinal canal and compressing the spinal cord (Fig. 1A-C). However, this was not observed on MRI 4 years prior (Fig. 1D). Retro-odontoid pseudotumor (ROP) grew in size during the 4 years (Fig. 1A and B). A solid small mass with iso-signal intensity on the T2-weighted images was present adjacent to the ROP in the cyst. Before surgery, we considered the cystic mass to have originated from the ROP and be located outside the dura mater.

Since the spinal cord was severely compressed by the cystic mass located laterally, we considered that direct cystectomy was feasible to improve neurological symptoms. Given the advanced age of the patient and lack of atlantoaxial instability, we did not utilize spinal fusion for coexisting ROP. Surgical removal of the cyst was performed via partial laminectomy of the atlas. At first, we could not find the cystic mass from the surface view in the spinal canal (Fig. 2A). Using intraoperative ultrasonography, we noticed that the cyst was located in the thecal sac (Fig. 2B). The dura mater was incised just above the cyst and opened. After clear serous fluid drained out, a yellow solid mass was observed under the arachnoid membrane adjacent to the right lateral side of the spinal cord (Fig. 2C). When the cyst wall was incised, yellow mucinous material protruded. After debulking the cystic mass, small penetration was observed at the anterior wall of the thecal sac (Fig. 2E). We complete the surgery after closing the dural penetration of the anterior wall and the dural incision of the posterior wall. Histopathological examination revealed a multiloculated cyst-like structure, with hyperplasia of collagen fiber (Fig. 2E).
Intradural Synovial Cyst with Retro-odontoid Pseudotumor

Fig. 2  Intraoperative views of the intradural synovial cyst (A, C) and a finding of intraoperative ultrasonography (B). Histopathological image of the cyst (hematoxylin and eosin stain, ×100, scale bar 200 μm) (D).

A: Location of the cystic mass was pointed out by the arrow, but the mass was not recognized from the view of the surface after partial laminectomy of the atlas.

B: Intraoperative ultrasonography showed that the cyst (arrowhead) was located on the right side of the spinal cord (asterisk) in the dura mater.

C: After opening the cyst, a yellow solid mass (arrowhead) was observed on the right lateral side of the spinal cord.

D: After debulking the cystic mass, the cyst wall was dissected around the whole surface.

E: Small penetration was identified at the anterior wall of the thecal sac after removal of the cystic mass.

F: Histopathological examination revealed a multiloculated cyst-like structure, with hyperplasia of collagen fiber (arrow), infiltration of inflammatory cells such as lymphocytes and macrophage (arrowhead), and growth of capillaries.

Discussion

We herein report a very rare case of IDSC accompanied by ROP. To the best of our knowledge, only 13 cases of intradural synovial cysts or ganglion cysts in the spinal canal have been reported previously (Table 1). Postoperative MRI showed adequate decompression of the spinal cord (Fig. 3A and B). The neurological symptoms were remarkably improved postoperatively, and the patient was able to perform normal activities of daily living after 2 months. Recurrence of the cyst was not observed until the latest follow-up after 3 years, during which the ROP decreased in size (Fig. 3C and D). Atlantoaxial instability or progressive spinal kyphosis was not observed in periodical plain radiographic studies.
was related to mechanical load on the thecal sac due to the tendon of rectus capitis. Although not pointed out, three of the five cases in the previous case reports showed atlantoaxial IDSC complicated with ROP on the sagittal image of MRI in the literatures. ROP was formed as noninflammatory masses due to ligamentous degeneration and...
tearing of the transverse ligament, followed by a cycle of attempted repair and mass formation. ROP is often associated with atlantoaxial instability but can be caused by atlantoaxial hypermobility, without radiological instability, secondary to a decrease of range of motion in the lower cervical spine. According to previous reports, the atlantoaxial synovial cyst is frequently formed on the transverse ligament and is also associated with atlantoaxial instability. In the present case, IDSC and ROP were caused by excessive loading and microinstability at the atlantoaxial segment due to a decrease in the range of motion at the lower cervical spine.

In the present case, preoperative diagnosis of the intradural location was quite difficult. Most IDSCs in the previous reports were located in the middle of the spinal canal, and the anterior boundary of the thecal sac was clearly observed. In the present case, however, IDSC occurred adjacent to the preexisting ROP on the lateral side of the spinal canal. Due to severe compression of the spinal cord, the boundary of the thecal sac could not be observed, resulting in misdiagnosis as an extradural lesion. Some authors noted that CT myelography may be useful for the accurate preoperative diagnosis of such a case.

In previous literatures, IDSCs have been commonly treated with direct cystectomy through C1 laminectomy. As IDSC occupies an intradural space, direct cystectomy is essential for immediate neural decompression. Recurrence or deterioration of the instability has not been reported. There was only one case report utilizing spinal fusion without cystectomy for the surgical treatment of IDSC with spinal instability due to rheumatoid arthritis. In this case, the preoperative symptom was dysesthesia without motor disturbance and the preoperative MRI showed that spinal cord compression was not severe. The authors decided to perform the posterior fusion for atlantoaxial instability prior to direct cystectomy and unexpectedly obtained shrinkage of the IDSC. According to the latest studies, ROP accompanying cyst formation is highly related to atlantoaxial instability, and regression of both ROP and the cyst can be obtained after posterior fusion. Although there is no description of an intradural cyst, it is possible that these studies included an IDSC mimicking an extradural lesion like the present case. The present case underwent direct cystectomy through C1 laminectomy. Recurrence had not been observed until the latest follow-up 3 years after the surgery, during which the ROP decreased in size. Unless obvious atlantoaxial instability is present, fusion surgery seems to be unnecessary. However, to confirm this conjecture, further studies on long-term surgical outcomes are necessary.

**Conclusion**

We experienced a rare case involving an IDSC associated with an ROP. For accurate preoperative diagnosis, the intradural location of a cystic lesion at the atlantoaxial level should be considered.

**Conflicts of Interest Disclosure**

The authors declare that there are no conflicts of interest. All authors who are members of the Japan Neurosurgical Society (JNS) have registered online Self-reported COI Disclosure Statement Forms through the website for JNS members.

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